EFFECTS ON SLEEP-STATE ORGANISATION OF A BEHAVIOURAL INTERVENTION FOR INFANT SLEEP DISTURBANCE

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Abstract

Establishing healthy sleep-wake patterns early in infancy is vitally important as sleep problems can persist. Behavioural sleep interventions such as the parental presence procedure are well established and have been found to improve infant sleep as determined by parent report. The exact nature of this improvement is, however, unclear. Sleep consolidation, sleep-state organisation, and self-soothing are thought likely to change after intervention; however, no known research has comprehensively determined which of these variables change as infant sleep changes in response to intervention. Three participants aged between 7 to 11 months who met the criteria for Infant Sleep Disturbance (ISD) were referred by a Health Centre and the parental presence behavioural sleep intervention was implemented. Parental report and videosomonography (VSG) data were used to measure sleep before and after intervention. While parental report is limited in that parents can only report what they can hear and/or see, VSG offers a tool that can be used to measure sleep-state organisation, state changes, and periods when the infant is awake and quiet. The present research found that infants' sleep became more consolidated resulting in fewer sleep-wake transitions and night wakings. Infants who had difficulties initiating sleep on their own also demonstrated decrease in Sleep Onset Delay (SOD). Furthermore, infants were found to sleep through a greater number of sleep-state transitions and sleep for a greater duration of time before waking. Collectively this research provides some evidence that changing parental behaviours to those that promote self-initiation through self-soothing and consistency, can change sleep-state organisation and improve self-soothing.

Within the first year of life, rapid change occurs in all aspects of infant development. One aspect that is of particular concern to professionals is sleep-wake organisation. This is of concern given that there is evidence that sleep problems can continue into adolescence and beyond (Dahl, 1996), lead to disruption in day-time functioning (Burham, Gaylor & Anders, 2006), affect physical growth (Lampl & Johnson, 2011; Tikotzky, Marcas, Har-Toov, Dollberg, Bar-Haim & Sadeh, 2010), and later impact on parental (Karraker & Young, 2007) and individual mental health (Gregory & O'Connor, 2002; Touchette, Petit, Seguin, Boivin, Tremblay & Montplaisir, 2007). Thus, establishing healthy sleep-wake patterns early in infancy is vitally important (Sadeh, 1994; Sadeh, Mindell, Luedtke & Wiegand, 2009; Selim, France, Blampied & Liberty, 2006).

The majority of typically developing infants over four months of age are able to sleep through the night without regular interventions from their parents (Henderson, France, Owens & Blampied, 2010), but about a quarter are not achieving settled sleep (Scott & Richards, 1990). In general, non-consolidated sleep before four months of age is consistent with typical sleep development and not considered problematic (Henderson et al., 2010) while problems such as Sleep Onset Delay (SOD) and recurrent night wakings after six months of age may be considered evidence of Infant Sleep Disturbance (ISD). After 24 months of age children have typically developed adequate language skills. This means that interventions are recommended which utilise their communication abilities and which are different from those used with younger children. For these reasons, the present literature review has, where possible, concentrated on infants between 4 to 24 months of age. Occasionally, research outside of this range is discussed, in particular in those areas where infant research is limited.

A sleep disturbed infant displays one or more of the following: bed-time resistance, SOD and/or recurrent night wakings (Blampied & Bootzin, 2013). A sleep disturbed infant is likely to sleep less, cry more, have difficulty self-soothing and may have a more difficult temperament during the day (France & Blampied, 1999). Other difficulties can manifest as night fears and parasomnias (Owens, France, & Wiggs, 1999), but these are not reviewed in the current study.

Research into infant sleep overwhelmingly suggests that behavioural interventions improve infant sleep, resulting in less distress and crying, and greater Sleep Efficiency (proportion of time spent asleep while in bed) (France & Hudson, 1993; France & Blampied, 2005; Hiscock & Wake, 2002; Sadeh 1990, as cited in Sadeh & Anders 1993; Sadeh, 1994; Sadeh et al., 2009; Owens et al., 1999). What is unclear is whether behavioural sleep interventions are changing infant's self-soothing behaviours (i.e., demonstrated by waking but remaining quiet) rather than signalling (i.e., crying upon waking) to their parents; or whether they consolidate sleep by increasing time asleep and decreasing sleep-wake transitions. This is an important question as it addresses the role of parent-child interactions on sleep. In other words, does changing an infant's sleep environment by reducing parent interactions at night impact on sleep-state organisation by increasing time asleep or, on the other hand does it promote self-soothing skills meaning the child continues to wake, while remaining quiet, but settles back to sleep of his or her own accord, or do both outcomes occur? To understand this question, it is necessary to first understand what is broadly meant by environmental influences. Furthermore it is important to be familiar with sleep terms such as 'sleep-state organisation' and 'self-soothing'. These concepts are discussed in more detail below. This chapter then reviews the current sleep research literature to illustrate the importance of the research question 'To what extent is sleep-state organisation influenced by environmental factors such as a behavioural intervention, and what is the nature of this change'?

To begin, an overview of the importance of infant sleep is discussed. The development of infant sleep and important sleep terms are then explained with particular reference to those used within the present research. The literature review then outlines the role of environment on sleep. After outlining key background material the literature relating specifically to the research questions, including, eitology of ISD, development of ISD, interventions to treat ISD, and tools commonly used to measure change after intervention is systematically reviewed.

The Importance of Sleep

Sleep disruption at night is associated with disruption in day-time functioning (Burham et al., 2006). Research also suggests that disruptive night waking in childhood is more prevalent in children who have had earlier sleep problems when compared to children who have had no history of sleep difficulties (Thorne & Skuladottir, 2006; Touchette, Petit, Paquet, Boivin, Japel, Tremblay & Montplaisir, 2005). What is unclear is whether sleep disruption at night has any long-term effects. This has been an area of particular research interest.

Sleep has many functions. It can influence brain functioning and development, human physiology and growth, ongoing sleep behaviours and routines, and may contribute to individual or parental psychopathology (Dahl, 1996). Determining causal relationships between infant sleep and other variables is a difficult task as the relationships between sleep and development are often complex and bidirectional in nature. Particular care should be applied when investigating the long-term developmental effects of sleep disturbance on the growing child and its effects into adolescence and beyond. At present, research in this area is variable and often costly when applying longitudinal research designs.

Brain Functioning and Cognitive Development

In general, researchers have found that sleep often increases following a learning task and that when sleep deprivation occurs it often impairs task acquisition and consolidation (Ednick, Cohen, McPhail, Beebe, Simakajornboon & Amin, 2009). Scher (2004) administered the Bayley Scales of Infant Development to fifty ten-month old infants and found that there was a small but significant correlation between the quality of infant sleep and cognitive achievement. Specifically, the more fragmented the infants' sleep (i.e., greater number of sleep-wake transitions) the lower the scores on the Mental Developmental Index (MDI). Scher (2004) also investigated the effects of sleep on psychomotor performance and found no significant associations. Other studies have found a link between brain plasticity and sleep deprivation. Brain plasticity refers to changes in brain structure and function in response to the environment. Dang-Vu and colleagues found that sleep deprivation impacts on an individual's ability to learn (Dang-Vu, Desseilles, Peigneux & Maquet, 2006). Brain plasticity decreases with age and therefore is heavily influenced during infancy when the individual is learning a great deal about their world.

Another fundamental aspect of sleep is its division into Active and Quiet Sleep. Active Sleep (or Rapid Eye Movement, REM Sleep), and Quiet Sleep (Non-Rapid Eye Movement, NREM Sleep) are terms used to describe two states of sleep. The role of each state is of curiosity to researchers. Dang-Vu and colleagues reviewed current sleep-state literature and concluded that Active and Quiet Sleep states are both important for brain development (Dang-Vu et al., 2006). Another study investigated the effects of a phenomena referred to as REM bursts (Becker & Thoman, 1981). REM bursts occur during Active (REM) Sleep and often have a seizure-like appearance. During this time an infant is likely to present with intense eye movements which can be accompanied by facial movements (i.e., brow raising or eye opening) and give a clinical impression of instability in the central nervous system. Becker and Thoman (1981) found that REM bursts at six months of age have a negative correlation with scores on the Bayley Scales of Infant Development at one year of age. Researchers concluded that REM bursts at six months of age may indicate some dysfunction or delay in sleep-state organisation which may impact later development. Conversely, a reduction of REM bursts is indicative of a maturation of the sleep-state.

Beckwith and Parmelee (1986) applied a longitudinal design to measure the effect of sleep-state on later development of pre-term infants. Trace alternant (high voltage slow waves) occur during Quiet Sleep and state transitions. Those who had a greater proportion of trace alternant during Quiet Sleep scored significantly higher on developmental assessments. The results suggest that trace alternant during Quiet Sleep may in part contribute to later cognitive development; however, there are some limitations of this study. Firstly, it was conducted on pre-term infants and therefore cannot be generalised to full-term infant development. Secondly, researchers only measured the first 60 minutes of a 90 minute sleep time. Research suggests that infants spend a greater length of time in Quiet Sleep during the first half of their sleep and therefore without assessing the full night of sleep it is difficult to establish a link between Quiet Sleep and cognitive development.

Taken together the above findings suggest that Active and Quiet Sleep both play an important role in healthy brain development and that disruption or deprivation of either sleep-state may contribute to long-term maladaptive outcomes. These studies may also suggest why Active Sleep is more prevalent during infant sleep as this is a period where the human brain is described as having more plasticity.

Physical Growth

The consolidation of healthy sleep-wake patterns during infancy is an important developmental task that may impact on later growth and development (Tikotzky et al., 2010). Lampl and Johnson (2011) conducted a study to investigate whether sleep and infant growth (body length) were correlated. Researchers found that there was a positive correlation between the length of daily sleep (in hours) and body length. This relationship was also mirrored when analysing quantity of sleep bouts and growth. Some gender differences were noted, with researchers describing ‘growing boys’ as experiencing longer sleep bouts and ‘growing girls’ requiring more sleep bouts.

Other researchers have investigated similar domains for night sleep. Tikotzky and colleagues examined the relationship between sleep and physical growth during the first six months (Tikotzky et al., 2010). To do so, they used the measure Weight-to-Length Ratio (WLR) and analysed its relationship to different sleep variables (i.e. Sleep Efficiency, duration of sleep, and number of night wakings). Overall, Sleep Efficiency (percentage of time asleep across a night) correlated negatively with WLR measurements and positively with length readings, suggesting sleep plays an important role in growth. This sample represented families of mid to high SES and therefore may limit the findings. Furthermore, more boys were also sampled than girls and all children were first-born.

Individual and Parental Psychopathology

Generally, research has addressed the implications of infant sleep for both the parent and the child. For instance, Karraker and Young (2007) investigated the relationship between infants' sleep behaviours at six months of age and parents' depressive symptoms. They found that the rate of clinically significant depression scores was twice as high in mothers of chronically waking infants when compared with mothers whose infants did not wake during the night (Karraker & Young, 2007) This finding could suggest that sleep deprivation caused by caring for a poorly sleeping infant could increase a mother's chance of developing clinically significant depressive symptoms. Another possible explanation is that mother's depressive symptoms influence the way they attend to their child and that it is the interaction between the parent and child that is influencing the infant's sleep difficulties.

One study investigated the long-term effects of sleep disturbance on the infant and concluded that sleep problems at four years of age may suggest an increased likelihood of depression or anxiety, inattention or over-activity, and aggression in early childhood and adolescence (Gregory & O'Connor, 2002). Other researchers have found similar relationships. For instance, Touchette and colleagues predicted that shorter sleep duration would significantly increase a child's risk for day-time externalising problems and lower cognitive performance (Touchette et al., 2007). Their findings supported this prediction as they found that short nocturnal sleep before the age of three and a half increased (2.5 times) the child's risk for high hyperactivity-impulsivity scores at six years of age when compared to children who slept for 11 hours a night.

Temperament and Behaviour

Infant temperament has been suggested as being one of the modulators of sleep-wake regulation (Scher, Tirosh & Lavie, 1998). Research in this area has identified a link between sleep difficulties and specific temperament profiles (Scher et al., 1998). The nature of this relationship is unknown as different temperament dimensions are highlighted in each study and the relationship has been described as both a possible contributor to sleep disturbance and a possible outcome. In other words, infant temperament may impact on their sleep, or infant sleep could impact on their temperament. Alternatively, both could be impacting on each other. General consensus is that the two are not mutually exclusive and rather impact on each other in a transactional manner.

Sadeh, Lavie and Scher (1994) reported that infants with a difficult temperament slept less than children with an easy temperament, while Weissbluth (1982), reported that infants with severe night waking problems may be at greater risk for developing difficult temperament. Spruyt, Aitken, So, Charlton, Adamson and Horne (2008) replicated similar findings. Spruyt et al (2008) found that an increase in sleep duration was positively correlated with more favourable temperament qualities; approachability (nature of initial responses to new stimuli), adaptability (the ease with which reactions to stimuli are modified in an adaptive way), and rhymicity (the regularity of physiologic functions). Similar findings have also been replicated for adolescents where difficult temperaments correlate with unhealthy sleep patterns (Moore, Slane, Mindell, Burt & Klump, 2010).

Co-sleeping is defined by Stein, Colarusso, McKenna and Powers (2001) as the practice of an infant sharing a bed with their parent/s. A study conducted by Kelmanson (2004) found that infants with a more difficult temperament, who required rocking or rhythmic movements to get to sleep, were more likely to co-sleep (or bed share with parent/s), slept too little, and moved a lot during their sleep. In comparison rhythmic infants were more likely to be ready for bed, less likely to resist, and slept more. This data was largely measured by parental report and therefore could be a reflection of parental cognitions rather than a direct relationship between temperament and sleep. Scher and colleagues conducted a study to investigate whether parental report within this area of sleep research is a valid measure (Scher, Epstein, Sadeh, Tirosh & Lavie, 1992). They applied both subjective (parental report) and objective (actigraph) measures to monitor sleep and compared the two. They found that parental report was subjective in that it appeared to be highly influenced by parental cognitions. Parents also completed an infant temperament questionnaire. When researchers examined the relationship between sleep and infant temperament they concluded there was a moderate correlation but noted that this may be a function of parental cognitions rather than a pure relationship between infant temperament and sleep.

Development of Sleep: Birth-24 Months

Sleep changes considerably in the first two years of life. Sleep-state organisation, sleep duration, Longest Sleep Period (LSP), Longest Self-Regulated Sleep Period (LSRSP) and night wakings are some measurable aspects of sleep that exhibit changes over time.

Sleep-State Organisation

As read above; infant sleep is organised broadly in two distinct sleep states, namely Active Sleep (later in development called Rapid Eye Movement/REM Sleep), and Quiet Sleep (later known as Non-Rapid Eye Movement/NREM Sleep). Active Sleep is characterised by uneven respiration, sporadic movements with low muscle tone between movements, and flickering eyelids indicative of eye movements (Anders & Keener, 1985; Halpern, MacLean & Baumeister, 1995; Thoman, 1990). Other infant sleep behaviours consistent with that of an Active Sleep state include, smiling, frowning, sucking, sighs, opening the eyes, twitching, and stretching (Thoman, 1990). During Quiet Sleep an infant has relatively slow and regular respiration (Anders & Keener, 1985; Halpern et al., 1995; Thoman, 1990). Motor activity is sparse (tonic muscle tone) and if present, usually consists of occasional startles, sighs, and rhythmic mouthing (Thoman, 1990). Generally Active Sleep clusters toward the end of the night and Quiet Sleep clusters toward the beginning (Halpern et al., 1995).

Furthermore, after a period of wakefulness an infant typically transitions into Active Sleep (Thoman, 1990). The duration of each sleep cycle within infancy is approximately 50 to 60 minutes (Roffwarg, Muzio & Dement, 1966). The period in between each sleep state is likely to involve mixed signs of both Active and Quiet Sleep (Thoman, 1990).

As an infant grows and develops quite considerable changes occur in the distribution of Active and Quiet Sleep. Specifically, the proportion of Active Sleep decreases and Quiet Sleep increases (Halpern et al., 1995; Parmelee, Schulte, Akiyama, Weener, Schultz & Stern, 1968). In other words, there is proportionally more Active/REM Sleep exhibited during infant sleep in comparison to the sleep of older children, adolescents, and adults (France & Blampied, 1999). Infants therefore demonstrate a progressive organisation of their sleep states (Halpern et al., 1995) and changes over time in the distribution of sleep periods between sleep states. At term, Active Sleep consumes between 47 to 50% of total sleep time, but by six months of age this has reduced to between 29 to 49% (Anders & Keener, 1985). Conversely, Quiet Sleep constitutes around 19 to 26% at birth, 40 to 42% at six months, and 46% at one year of age (Anders & Keener, 1985). Other researchers have found similar results (Parmelee et al., 1968).

Sleep Duration

Age is one of the most important early developmental factors contributing to sleep duration (Thorleifsdottir, Bjornsson, Benediktsdottir, Gislason & Kristbjarnarson, 2002). At birth, most full-term babies will sleep intermittently for approximately 16 to 18 hours during a 24 hour period (Sheldon, 2006; Touchette, Petit, Tremblay & Montplaisir, 2009). The duration of sleep within each day declines as the infant develops (Burham et al., 2006; Touchette et al., 2009) so that by 16 weeks of age an infant will typically sleep for 14 to 15 hours a night and by six to eight months of age this decreases to 13 to 14 hours a night (Sheldon, 2006).

The nature of sleep in newborns consists of multiple sleep periods within a 24-hour period; typically an infant will alternate with three to four hour sleep periods followed by one to two hours of wakefulness (Mindell & Owens, 2009). As an infant develops, their sleep becomes more consolidated as it also becomes entrained by a circadian rhythm (Salzarulo & Ficca, 2002). This means that a longer duration of sleep should occur at night and a shorter duration of sleep during the day. On average, this night-time consolidation occurs by three months of age (Henderson et al., 2010). Evidence of this process is shown in the development of the Longest Sleep Period (LSP).

Longest Sleep Period (LSP)

The Longest Sleep Period (LSP) is a sensitive measure used to approximate infant sleep-wake organisation by measuring the infant's physiological capacity for continuous sleep (Anders, Keener, Bowe & Shoaff, 1983; Henderson et al., 2010). To measure this, each sleep episode is recorded beginning when the infant falls asleep and stopping when the infant reaches wakefulness to end that episode (Anders et al., 1983; Anders & Keener, 1985; Henderson et al., 2010). Over a night with several sleep episodes, one will be the LSP. During infancy, the LSP typically increases with age. Sleep periods lengthen considerably during the first few months of life from approximately four hours at two weeks of age to seven hours by approximately five months of age (Anders & Keener, 1985). As part of circadian entrainment, by three months of age the sleep-wake cycle is consolidated so that the LSP occurs during night and the longest wake period occurs during the day (Sheldon, 2006). Hall and colleagues even found that there was a significant increase in LSP after parents received psychoeducation on infant sleep and management (Hall, Saunders, Clauson, Carty & Janssen, 2006). Henderson and colleagues (2010) also propose another measure, Longest Self-Regulated Sleep Period (LSRSP) for sleep-state organisation that includes quiet wakefulness.

Longest Self-Regulated Sleep Period (LSRSP)

While its application to sleep research is relatively new, LSRSP offers a quantitative measure of sleep-state organisation that encompasses an important aspect of infant sleep, which is that of the infant developing the skills to reinitiate sleep on their own (Henderson et al., 2010). To calculate LSRSP researchers combine the total period of sustained sleep with consecutive periods of quiet wakefulness and sleep resumption. When this measure is applied over time it may provide valuable information regarding the consolidation of sleep. In other words, an increase in LSRSP would indicate sleep maturity as it encompasses times of quiet awake or self-soothing which are particularly relevant to the present research as this would indicate a progression away from parental soothing at night in favour of self-soothing. Alternatively, measuring LSP and duration of quiet awake would provide sufficient data to assume this also.

Night Wakings

During early childhood the most common complaints about infant sleep reported by parents are to do with excessive night waking and difficulties with sleep initiation (Sadeh et al., 2009). Night wakings are defined as wakings that are coupled with any type of signalling, i.e., crying, calling out, or any other signal that may attract the parent's attention (France & Blampied, 1999). These are the wakings that parents often report and respond to (i.e., by feeding, cuddling, or changing). Little is known about infant wakings that are quiet (i.e., without crying, fussing, or talking) as these are not reported, given parents cannot hear the waking, although they are detected by some methods such as videosomonography (VSG).

As a consequence of repeated cycling between Active and Quiet Sleep, most infants wake during the night (Ferber, 1985). Some of these wakings are signalled while others are associated with a period of quiet wakefulness. Those that are signalled may attract parental attention and nurturant care, while those that are not signalled typically escape parental attention, and the child self-regulates their return to sleep, as measured by the LSRSP. Signalled waking (and other sleep problems such as initial difficulty in settling) may become persistent and if left untreated, such ISDs can become a chronic problem (Sadeh, 1994).

It is however, important to note that wakings from sleep are frequent during the first two months of life and therefore are typical of early infant development (Henderson et al., 2010). From about two to four months of age most infants should be able to sleep through the night without regular interventions from their parents, however if night waking becomes persistent and meets the criteria stated above, sleep disturbance may be considered as a clinically significant problem (Henderson et al., 2010). Anders, Halpern and Hua (1992) found in a sample of normal full-term infants that, all infants aroused from sleep at least once during the night at both three weeks of age and three months of age. When participants were tested at three weeks of age, 90% signalled that they were awake upon arousal. Conversely, when tested at eight months of age, 52% (or 11 infants) did not signal, and seven of the nine infants who did signal were defined by their parents as 'problem sleepers'. This result suggests that by eight months of age the infants in this study were waking and not signalling to their parents proportionally more often than when they were three weeks of age. This could suggest a developmental period where infants develop self-soothing skills that aid in managing their own arousals without parental involvement.

These developmental changes emphasise the importance of understanding the terms, 'sleeping through' and 'self-soothing'. As all infants frequently wake when transitioning between Active and Quiet Sleep and may either signal or not during the awake period (see above), the term 'sleeping through' does not necessarily mean sleeping without waking (Henderson, France & Blampied, 2011). In fact, infants without sleep problems may regularly wake and then self-initiate sleep following a night waking (Anders et al., 1992). Sleeping through therefore implies sleeping without parental intervention which is measured by the LSRSP. To achieve this, an infant requires the capacity to independently reinitiate sleep following nocturnal arousals, and this is referred to as self-soothing (Henderson et al., 2011). A further summary term to use when assessing the quality or quantity of sleep is Sleep Efficiency whereby the percentage of time spent asleep whilst in the basinet/cot is used for measuring changes in sleep after intervention (Coons & Guilleminault, 1984; Scher, Epstein, & Tirosh, 2004). The equation for Sleep Efficiency is (total time asleep/total time available for sleep) multiplied by 100. An important question introduced below is the role of environment on sleep.

Environmental Influences

Circadian Rhythm and Zeitgebers

One indication that environmental factors influence sleep comes from what we know of the circadian entrainment process, a process which leads to systematic changes in the distribution of sleep across night and day. Humans, along with many other animals, are adapted to adhere to a circadian rhythm which consists of sleeping during the night and being wakeful during the day (Moore, 2006). Sleeping during the night provides optimal time for restoration and recovery, while being wakeful during the day ensures optimal use of light to aid vision (Moore, 2006). Circadian rhythms develop during childhood within a brain structure called the Suprachiasmatic Nuclei of the Hypothalamus (Gachon, Nagoshi, Brown, Ripperger & Schibler, 2004). While humans have an internal circadian timing system (Emens, Lewy, Lefler & Sack, 2005) with a time of approximately 25 hours, this is reduced to 24 hours when entrained by the external day-night cycle (Mitru, Millrood & Mateika, 2002). In other words, we rely on external environmental factors to inform us of time (i.e., light, clocks, alarms, temperature). These factors are known as Zeitgebers and without them our sleep and behavioural patterns would differ from the 24-hour cycle that we know. For example, a study conducted by Figueiro and Rea (2010) found that adolescents had a later sleep onset and slept for a shorter duration during spring than in winter. This suggests that external factors such as light can change sleep patterns.

Light has also been implemented as an effective intervention for improving sleep for children and adolescents (Fromm, Horlebein, Meergans, Niesner & Randler, 2011), and adults who work shift hours (Eastman, Boulos, Terman, Campbell, Dijk & Lewy, 1995). For instance, Fromm et al (2010) investigated the effects of a Wake Up Light (WUL) on subjective wellbeing and wakefulness in a sample of children and adolescents (Fromm et al., 2011). Researchers found that the WUP resulted in significant changes in reported alertness and willingness to wake. Other studies have investigated sleep effects on blind or visually impaired individuals and found that other factors such as age may play a larger role in sleep disruption rather than external visual cues (Lazreg, Laatiri & Dogui, 2011).

Family Environment and Sleep Hygiene

Family environment and parent interactions have also been found to impact infant sleep. For instance, disorganised home environments including poor sleep hygiene have been linked to sleep disturbance in infants (Gregory, Eley, O'Connor, Rijsdijk & Plomin, 2005) and school aged children (Biggs, Lushington, Van den Heuvel, James Martin & Declan Kennedy, 2011). Conversely, intervention involving more organised sleep routines has been found to improve sleep in infants (Gabrielle, Grote & Jonas, 1981) adolescents (Billows, Gradisar, Dohnt, Johnston, McCappin & Hudson, 2009), adults (Lee & Gay, 2011), and the elderly (Stepanski & Wyatt, 2003). Sleep hygiene refers to behaviours and routines that precede sleep. While researchers have found a relationship between sleep hygiene and sleep disturbance, many highlight a strong need for further research (Biggs et al., 2011; Billows et al., 2009; Zisberg, Gur-Yaish & Shochat, 2010).

Inconsistent bed-time routines may contribute to ISD. A bed-time routine is a relatively simple behavioural intervention that can be implemented easily (Adams & Rickert, 1989). The rationale for its benefit is that daily routines in general lead to predictable and less stressful environments (Mindell, Telofski, Wiegand & Kurtz, 2009). A bed-time routine involves consistent and predictable behaviours leading up to the infant being placed in their basinet or cot (Mindell et al., 2009). The same activities therefore take place in the same order each night. Mindell et al (2009) found significant reductions in problematic sleep behaviours (i.e., sleep onset delay, frequency of night wakings, and duration of night wakings) after the implementation of a consistent bed-time routine when compared to controls. Interventions of this nature have also been used in settings such as hospitals as a therapeutic intervention for adults (Gardner, Collins, Osborne, Henderson & Eastwood, 2009).

Twin studies allow researchers an opportunity to estimate the contribution of genes and environment (Barclay, Eley, Buysse, Rijsdijk & Gregory, 2010). To do so, researchers closely examine the similarities and differences between identical twins who are raised in different environments (Barclay et al., 2010). This way it is possible to estimate the relative contribution of heritability and environmental influences on factors of behaviour such as infant sleep. Research within this area suggests that in fact both genes and environment play an important role in sleep (Barclay et al., 2010; Barclay, Eley, Rijsdik, & Gregory, 2011; Moore et al., 2006), although one study found that sleep characteristics were predominately influenced by environmental factors (Brescianini et al., 2010). Brescianini and colleagues studied 314 18-month old twins (127 monozygotic and 187 dizygotic) by means of parental report and found the following sleep characteristics to be linked to environmental factors: co-sleeping (98.3%), sleep duration (64.1%), and waking episodes of at least seven per week (63.2%). In summary some of these studies point to the role of the environment in shaping sleep behaviours, it is therefore important to investigate more comprehensively the role environment in terms of parent-child behaviours and its role in the etiology and development of ISD.

Etiology

Infant Sleep Disturbances (ISD) have largely been attributed to sleep-state organisation factors, poor infant self-soothing skills, excessive involvement from either or both parents, and sleep hygiene (Sadeh et al., 2009). In particular, much research attention has focused on the behavioural aspects of ISD, more specifically, the impact of parental behaviour on infant sleep. Parental responses to night wakings are well documented in current literature.

One study investigated this extensively by administering questionnaires to parents of infants aged 13 months to assess what settling strategies were most common and to investigate their role in infant sleep behaviours (Morrell & Cortina-Borja, 2002). Results revealed that the strategies most commonly used by parents to settle their infants to sleep were giving a feed, talking softly to the child, cuddling in the arms, and stroking. Five main factors were used for describing the settling strategies commonly used by parents, namely, active physical comforting (i.e., cuddling), encouraging infant autonomy (i.e., leaving to cry), movement (i.e., car rides), passive physical (i.e., standing next to cot without picking up the infant), and social comforting (i.e., stories). Morrell and Cortina-Borja (2002) concluded that parents who excessively used active physical comforting as a strategy when combined with limited encouragement of autonomy strategies were more likely to have infants with sleeping problems.

Touchette and colleagues also found that parental behavioural responses at bed-time in response to nocturnal wakings are highly associated with an infants' sleep-state consolidation (Touchette, et al., 2005). Researchers found that the parental responses most strongly associated with less than six hours of consecutive infant sleep were putting the infant to bed asleep (or staying with them until asleep), feeding infants after wakings, rocking infants or placing them in their parents' bed after wakings, difficult temperament, and co-sleeping (sharing a room or bed with parents or siblings) (Touchette et al., 2005). Research suggests that all of these responses act as reinforcement for the wakings, and hence create a coercive cycle between the parent and infant (Blampied & France, 1993). Based on this, researchers have concluded that early intervention could possibly break this cycle by intervening at the environmental level by changing parent interaction (France & Hudson, 1990), specifically by eliminating or at least reducing parental attention to non-reinforcing levels, that is the behavioural procedure called extinction.

Some research suggests that parental cognitions can act as a barrier for changing parent interaction at night-time. For instance, Sadeh, Flint-Ofir, Tirosh & Tikotzky (2007) investigated what role parental cognitions play in the development of ISD, and in particular their role in parental responses such as those investigated by earlier researchers. Sadeh et al (2007) found that both mothers and fathers of infants with ISD reported difficulties with setting limits (resisting infants' demands). Interestingly when these parents were presented with hypothetical cases of infants with sleep difficulties they reported limit setting to be an appropriate approach to promote sleep consolidation for the infants. This finding suggests that parental cognitions may act as a barrier to implementing behavioural strategies such as limit setting.

Parents who implement limit setting behaviours such as withdrawing reinforcement for signalling upon awakening have been found to promote sleep consolidation in infants. For example, infants whose parents delayed their response to signalling (i.e., crying) at three months of age were more likely to soothe themselves to sleep at one year of age (Burham et al., 2006). Sadeh, Tikotzky & Scher (2010) later wrote that intervention based on modifying parental behaviours and cognitions can improve infant sleep, stating that parental personality, psychopathology, and cognitions all contribute to the way in which parents respond to their infants' nocturnal wakings and hence influence infant sleep (Sadeh et al., 2010). Sadeh and colleagues do, however, stress that these links appear bidirectional in that poor infant sleep may influence parental behaviours, and vice versa that a family stressor such as poor infant sleep may be a risk factor for maternal depression (Sadeh et al., 2010).

One solution parents may rely on when infants wake frequently during the night is to allow the infant to sleep in their bed (i.e., co-sleep; Sadeh & Anders, 1993). This is problematic as young children who co-sleep have been found to have a more disrupted bedtime routines and less consolidated sleep-wake patterns (Hayes, Parker, Sallinen & Davare, 2001). Parental beliefs about infant temperament have also been found to impact co-sleeping, for example, mothers who have young children who co-sleep also report their children as having more difficult temperaments (Hayes et al., 2001; Kelmanson, 2004; Scher et al., 1992).

Although co-sleeping between parent and child is not common practice in Western cultures, in certain societies co-sleeping in infancy is normal practice and is believed to enhance the parent-child attachment and infant security (Oppenheim, 1998; Sadeh & Anders, 1993; Welles-Nystrom, 2005). In Western society the impact of co-sleeping on the development of healthy sleep- state organisation is controversial. For instance, Hunsley and Thoman (2002) found that sleep-state organisation differed in infants who co-slept from that of controls. Quiet Sleep increased and researchers speculated that this pattern could indicate that co-sleeping negatively impacts neurobehavioural development as infants spend less time in Active Sleep. Conversely other researchers have found no negative effects of co-sleeping (Okami, Weisner & Olmstead, 2002).

One possible interpretation of these contradictory findings is that disturbed sleep is not an outcome of co-sleeping per se, but rather caused by inconsistent sleeping routines and behaviours. For example, co-sleeping is often highly correlated with other parental behaviours such as frequent breastfeeding (Ball, 2003; Hayes, Roberts & Stowe, 1996; Taylor, Donovan & Leavitt, 2008). Taylor et al (2008) found that the frequency of co-sleeping per week was positively associated with frequency of breastfeeding. In other words, those infants who co-slept were also likely to breastfeed frequently during the night. Ball (2003) even concluded that in some instances co-sleeping can promote breastfeeding during the night. Other researchers have found that breastfeeding, night feedings in the parent's bed, and returning to sleep in the parent's bed were precursors to co-sleeping later in early childhood (Hayes et al., 1996).

Collectively, these studies suggest that parental behaviours and night-time activities impact on infant sleep, and that night sleep reciprocally impacts on parent behaviours. In summary, a number of possible factors may influence whether an infant develops disruptive or healthy sleep patterns. In fact, it is likely that the development of ISD is multifactorial and involves a much more complex model of development than any one factor discussed thus far. Models of how ISD develops have been proposed by Sadeh and Anders (1993), and Blampied and France (1993), and France and Blampied (1999).

The Development of Infant Sleep Disturbance

Two models are proposed as integrative ways of summarising infant sleep and its disruption; a family systems model (Sadeh and Anders, 1993) and a bio-developmental-behavioural model (Blampied & France, 1993; France and Blampied, 1999). Both models are discussed and offer a great deal when conceptualising ISD as they are compatible rather than competitive explanations of infant sleep and its difficulties.

Sadeh and Anders (1993)

Sadeh and Anders (1993) compartmentalise their model of ISD into two core elements, namely components, and outcomes. Components are factors which can influence an infant's sleep outcomes. The relationship between components and outcomes is considered transactional, meaning that the infants' symptoms are dynamic and can affect the whole family system; for example, sleep disturbance in infants can lead to family disruption, however, family disruption can also lead to ISD. In other words the relationship between the components of an infant's life and their sleep behaviour is bidirectional and reciprocal.

Proximal components include those that are close to the infant (i.e., parents), while distal components have a more remote influence (i.e., culture). Components can also be subdivided into the categories of extrinsic (factors outside of the infant) and intrinsic (factors within the infant). Combinations of proximal/distal and intrinsic/extrinsic are discussed below.

Distal extrinsic factors that may influence infant sleep (or be influenced by infant sleep) include cultural (social and cultural norms), environmental (economic pressures, caretaking), and family factors (family disruption or stress). These distal factors are considered as secondary influences on the infant as they more directly affect the familial context and parental behaviours. Distal factors can further influence (or be influenced by) extrinsic parental factors (personality, psychopathology, representations or internal working models) and/or intrinsic infant factors (health, developmental maturation, and temperament). The link between all the factors discussed is referred to as a mediating factor which acts as a mechanism linking the influencing components in an infant's life and the sleep outcome. Sadeh and Anders (1993) propose two main types of mediating factors, namely the attachment relationship between the parent and infant, and the interactive behaviours between the parent and infant (bed-time interactions, soothing behaviours, limit setting, co-sleeping). The literature reviewed thus far, has outlined how factors such as limit setting can influence the way parents respond to their infants nocturnal wakings and thus the infant can create learnt sleep behaviours (Sadeh et al., 2010).

Sadeh and Anders (1993) state that three key ways in which children can express sleep difficulties are by sleep onset problems, sleep continuity problems, and a mixture of the two. Sleep onset problems refer to difficulties falling asleep without assistance. Parents can sometimes respond to these difficulties by assisting in sleep onset by using techniques such as feeding, rocking, and cuddling. When using terms such as night wakings, Sadeh and Anders (1993) describe problematic infant wakings as those that are paired with infant signalling (i.e., crying) or difficulties self-soothing back to sleep. Unfortunately, parents are only aware of those wakings that are paired with infant signalling.

Sadeh and Anders (1993) recommend that assessment of infant sleep should be considered within the functioning of the entire family. Following the idea of a transactional and bidirectional model of infant sleep, successful intervention applied to any one area should result in widespread change within the entire system.

Blampied and France (1993) and France and Blampied (1999)

During the first six months of life the infant and his or her parents are confronted with the challenges and demands that result in the child either developing or failing to develop the skills needed to initiate sleep on their own i.e., to self-soothe. The outcome achieved during this stage creates future potential vulnerabilities beyond six months of age when the infant may develop primary sleep disturbance. Independent of the outcomes achieved during prior periods of infancy, disturbance may occur where an infant may revert back to regular waking after previously achieving a healthy sleeping regime, called secondary sleep disturbance. Importantly, the model outlines the antecedents and consequences associated with the bed-time schedule within each stage of development.

Self-soothing and sleep-initiation. During the first few months of life a child may be predisposed to ISD (France & Blampied, 1999). Whether the child has high or low constitutional or environmental vulnerability will in turn influence parental behaviour (France & Blampied, 1999). Parental responsiveness to crying will be in part determined by parents' own experiences of being parented, their beliefs (cognitions) about parenting, their personalities, and own psychopathologies (France & Blampied, 1999). This interactive relationship between the infants' own susceptibility and the parents' experiences will determine whether the parent responds appropriately to the infants' signals, or whether they will respond in an overly stimulating manner (France & Blampied, 1999), thereby, creating either appropriate or inappropriate reinforcement contingences for their infant's sleep-regulatory and sleep-disruptive responses. This is consistent with the earlier stated model by Sadeh and Anders (1993) in that bed-time routines within the environment (such as limit setting or co-sleeping) can influence infant sleep patterns.

Further, parents, by their behaviours in response to their infant, establish either appropriate or inappropriate proximal cues for sleep onset (France & Blampied, 1999). Appropriate cues would include placing the infant in their cot awake, and inappropriate cues can include activities such as rocking the infant to sleep or feeding them to initiate sleep (France & Blampied, 1999). Appropriate cues promote self-soothing and sleep-initiation. Inappropriate cues greatly hinder this process and result in sleep-initiation and/or self-soothing difficulties for the infant (France & Blampied, 1999) because the child learns to associate these parent-provided cues with sleep, and thus they require the same parental interactions each time they fall asleep. Both pathways create habits in which the outcome either results in parental-infant interaction being required for sleep initiation, or infant self-soothing so as to enter sleep on their own. If the child is put to bed awake and learns to initially fall asleep on their own then they are more likely to repeat this pattern if they wake during the night (Anders et al., 1992; Burnham et al., 2006; Henderson et al, 2010; Henderson et al, 2011).

Primary sleep disturbance. The interactions between the child and parent influence whether the infant will meet the next sleep milestone with or without self-soothing skills (France & Blampied, 1999). The need for regular waking in the night should decrease by three months of age as the LSP/LSRSP increases and thus so should the infants' nightly wakings (France & Blampied, 1999). If, during the night, the infant does not fully wake, then night-waking will not manifest itself (France & Blampied, 1999) Instead, the infant will sleep throughout the night either with or without a prior Sleep Onset Delay (SOD), depending on self-soothing skills developed over the first few months of life through experiences with sleep initiation (France & Blampied, 1999). If the infant wakes fully during night-time arousals one of two things may occur: either the infant re-initiates sleep without attention from their parents, or they will signal for parental attention through crying (France & Blampied, 1999). In other words, the model assumes that the way in which the infant behaves (i.e., with or without self-soothing to sleep) during sleep onset will mirror their reaction following an arousal from sleep later in the night and determine whether resumption of sleep requires parental interaction. For example, if the bed-time routine consists of rocking or feeding the infant to sleep at the start of the night then the same pattern of interactions will be required upon night-time wakings (also referred to as coercive behaviour trap as infant's duration of crying increases to signal parental attention and parental interaction increases upon infant distress). Conversely if parental interaction is reduced or withdrawn at bed-time (also referred to as extinction) then the infant may learn to self-soothe and reinitiate sleep on their own as they had during sleep onset. In summary, this means that the acquisition of self-soothing skills in the first few months of life is a protective factor for the development of primary sleep disturbance, and without these skills it is very likely that not only sleep onset delay develops but also primary sleep disturbance will emerge (France & Blampied, 1999).

Secondary sleep disturbance. It is also important to address the last part of France and Blampied's model, which is concerned with secondary sleep disturbance. Even infants who have settled to regular sleep patterns by six months of age are at risk of reverting to disrupted sleep patterns later in childhood (France & Blampied, 1999). This is what is known as secondary sleep disturbance, and is often sparked by a disruptive event such as child illness, birth of a sibling, family relocation, parental separation or other family disruption (France & Blampied, 1999).

Summary of the Models

In summary, both Sadeh and Ander's (1993), and France and Blampied's model of ISD illustrate how reciprocal and interactive environmental and social/personal factors contribute to infant sleep. While Sadeh and Anders explore possible environmental and individual contributors to ISD, France and Blampied focus more heavily on how these contributors may interact and influence sleep development over time through the behavioural process they entail. In combination, the two models illustrate comprehensively how ISD can be manifested. For instance, Sadeh and Anders (1993) illustrate effectively how the relationship between the parent and child is bidirectional, and highlight how factors within each interact and influence each other. Importantly Sadeh and Anders (1993) highlight the point that an improvement in one area system (child, parent, home, culture) can have a cascading effect whereby improvement results in the entire system. France and Blampied's model focuses more on how these same contributors develop over time, they discuss how an infant's vulnerability to sleep problems (such as sleep-state organisation or other aspects of temperament at birth), parental behaviour and disruptive events all appear to contribute to whether a child develops ISD. For instance, parents of sleep disturbed infants have a tendency to use varied and over-stimulating techniques to manage their child's ISD in comparison to parents of non-sleep-disturbed infants (France & Blampied, 1999). Such stimulating techniques include, but are not restricted to putting infants to bed asleep, remaining present with them as they fall asleep, sleeping with their child in their own bed, and feeding them during the night on wakings. In fact, positive results have been reported when these types of excessive and over-stimulating parental behaviours are reduced. As both models highlight, parental interaction (and the reciprocity between infant and parent) during bed-time routines and at night can mirror how an infant responds to night wakings (i.e., self-soothe or signal for parent).

Behavioural Interventions

As discussed above, during infants' first year of life they experience rapid changes in the consolidation of their sleep-wake patterns (Henderson et al., 2011). These changes are in part influenced by the infants' own physiological maturation, behavioural self-regulation and factors such as parental attention (Henderson et al., 2011). The type of treatment for sleep disturbance implemented therefore is dependent upon the interaction of these contributing factors. Interventions commonly used to treat infants with ISD range from pharmacological sedatives (such as Trimeprazine)[1] through to behavioural interventions (France & Hudson, 1993). Meijer (2011) states that most sleep disturbances appear to be maintained by learnt behaviour patterns. Of the behavioural interventions implemented for sleep disturbances in infants (aged 6 to 24 months), those which are most effective are found to encourage appropriate independence and self-soothing in the infant (Middlemiss, 2004; Sadeh et al., 2009). These types of outcomes are usually achieved by educating parents in sleep hygiene and teaching them skills to cope with sleep initiation disruptions or wakings during the night (Owens et al., 1999). For instance, Sadeh et al (2009) found that the most common factor underlying effective sleep interventions is the withdrawal of parent's excessive responses to sleep events. This is further supported by the bio-developmental-behavioural model discussed earlier

[1] Trimeprazine is no longer approved for use with infants

in that extinguishing or reducing parental interaction at night can facilitate the development of self-soothing and healthy sleep-wake patterns (France & Blampied, 1993; Blampied & France, 1999).

Parental attention could be removed altogether (extinction), however, this is often considered distressing to the child and the parent (France & Blampied, 2005) and may result in a post-extinction-response-burst (PERB), whereby the child increases the variability, frequency, and intensity of their response such as crying and calling out (France & Hudson, 1990). Modifications of interventions involving full withdrawal of parental attention seemed more appropriate to some investigators. One such alternative is systematic ignoring with minimal check procedure (Ferber, 1985) that involves the parent only checking their infant at set regular intervals while applying the extinction procedure stated above (France & Blampied, 1993; Blampied & France, 1999; France & Blampied, 2005). Hiscock and Wake (2002) conducted a study which employed a modified version of this intervention called 'controlled crying'. Parents were told to only respond to their infants cry at certain time intervals. These intervals were gradually increased so that the infant learnt how to self-soothe and fall asleep on their own. Researchers found significant improvements in infant sleep and maternal mood. It should be noted, however, that this procedure has some adverse features that require caution in its use (France & Blampied, 2005).

Another intervention is the parental presence procedure (Sadeh 1990, as cited in Sadeh & Anders 1993; Sadeh, 1994). The parental presence procedure is a short-term environmental and behavioural intervention that the parents implement (Sadeh 1990, as cited in Sadeh & Anders 1993; Sadeh, 1994). During the intervention parents are instructed to sleep in their infant's room. This is either initiated upon the infant delaying sleep onset or resumption, or when the parent is ready to settle to sleep for the night. Whilst the parent does not interact with the infant, their presence is thought to promote the infant's feelings of security while also encouraging the development of the self-soothing skills required to initiate sleep. The basis of the intervention was formulated predominately around theoretical underpinnings of attachment theory in that the presence of the attachment figure will increase feelings of security in the infant (Sadeh et al., 2010). The parental presence programme works in parallel with this theory by allowing the child to see there is no immediate danger upon waking, while at the same time preventing parental attention from reinforcing any waking and crying behaviour (France & Blampied, 2005; Selim et al., 2006).

The parental presence procedure is considered preferable to many other interventions as it attempts to counter concerns about infant distress that is speculated to occur in some of the other ISD behavioural interventions (France & Blampied, 2005). Middlemiss (2004) conducted a review of current sleep interventions and concluded that sleep interventions that promoted solitary sleep patterns and self-soothing were the most successful and that parental presence during the night can contribute to positive child development. Effectively the parental presence procedure combines both these aspects together.

The Effect of Intervention on Sleep-State Organisation

Environmental factors are considered one likely contributor to ISD (Sadeh & Anders, 1993) and its development over time (France & Blampied, 1993; 1999). Research investigating the effects of environment on infant sleep has further highlighted this role (Brescianini et al., 2010). Sadeh and Anders (1993) also described a cascading effect whereby improvement within one area results in the entire system change. As stated earlier the withdrawal of parent's excessive behaviours in relation to infant night-time disturbance has been found to be an effective environmental change and the most preferable behavioural intervention. In relation to Sadeh and Anders (1993) model this would be referred to as a proximal extrinsic factor. Behavioural interventions intervening within the parent-child environment are found to be effective in empirical studies (i.e., France & Blampied, 2005; France & Hudson, 1990; Gabriel et al, 1981; Hiscock & Wake, 2002; Sadeh, 1994; Selim et al., 2006). However, gaps arise in the research investigating the ongoing effects on infant sleep-state organisation in response to these interventions. This is because most of the phases in studies of this nature have relied on parental reports of their infant's sleep via daily sleep diaries.

Parent report is limited in so far that parents can only detect the wakings that are coupled with signalling (i.e., crying) (Sadeh et al., 2009). At present, it remains unclear as to whether this means the infant is going to sleep more promptly, sleeping for longer periods without waking (i.e., increasing the LSP), or that the infant is waking as often as before but managing their own arousals better and hence not signalling (i.e., increasing the LSRSP). As these are not incompatible developments different combinations of these outcomes may be shown by different infants, or by the same infant at different stages. Other researchers agree about the need to understand change in sleep-state organisation as related to intervention outcomes. To achieve this requires moving beyond parent report or sleep diaries and to use methods of assessment that capture changes in sleep-state organisation (Insana, Gozal & Montgomery-Downs, 2010; Sadeh, Acebo, Seife, Aytur & Carskadon, 1995; Sadeh, 2005).

With the benefits of such measurement, researchers can determine whether the behavioural interventions are changing sleep-state organisation or whether infants are learning to regulate their responses by self-soothing on awakening without necessarily increasing their time spent asleep. Before this is achieved, a measurement tool for accurately measuring both quiet and audible wakings as well as the other sleep variables previously discussed (i.e., LSP, LSRSP) should be investigated. Measures that contribute to this objective are Polysomnography (PSG), Actigraphy, and Videosomnography (VSG). These in combination with parental report are discussed and compared further below, outlining both strengths and limitations of each method.

Measuring Sleep

Polysomnography (PSG) has been considered as the (biological/physiological) 'gold standard' for identifying sleep-state changes (Insana et al., 2010). PSG involves a comprehensive recording of the bio-physiological changes that occur during sleep (Insana et al., 2010). It is generally used within a lab setting as the equipment is complex and involves various monitors recording brain activity, eye movements, muscle activity, skeletal muscle activation and heart rhythm during the night (Insana et al., 2010). The equipment while very reliable and valid has limitations in its application as it is expensive, time-consuming and intrusive so is not ideal for use on infants (Insana et al., 2010). It is difficult to use in the home environment and the necessity to relocate to the sleep lab may disrupt sleep-state organisation unless several nights are recorded for adaptation to the lab and the equipment. These factors limit its use in research of the kind undertaken in this study.

Actigraphy on the other hand can be used in any setting. The actigraph is a small, unobtrusive computerised activity monitor that detects movement when placed around the infant’s ankle. Actigraph recordings can differentiate when a child is asleep, awake, in Quiet Sleep or Active Sleep. Furthermore, it is cost-effective, permits monitoring for extended periods of time and is widely used in sleep studies (Sitnick, Goodlin-Jones & Anders, 2008). Actigraphy has been found to provide researchers with a method to determine valid representations of the sleep-wake patterns in infants (Wood, Kuntsi, Asherson & Saudino, 2008), however, some studies have questioned the validity of using actigraphy (Insana et al., 2010). One of the main concerns is that the actigraph is unable to detect sound and therefore it is not possible to distinguish between night wakings accompanied by signalling and those which are quiet.

Researchers have found that parental reports can differ quite dramatically from actigraphy data (Sadeh, 1994; Tsai, Blurr & Thomas, 2009). Sadeh (1994) investigated the sleep patterns of 50 infants with sleep disturbances using actigraph and parental reports during both baseline and intervention. When the actigraphy recordings were compared to actual parental reports, significant deviations were found although both parent report and actigraph recordings indicated an improvement in sleep during the intervention. Specifically, the number of night wakings reduced and

Sleep Efficiency increased. Although the two measures reported similar trends, there were some discrepancies in the actual values. Parents rated their children sleeping more when compared to the actigraph recordings and further reported a greater decrease in night-time waking than the actigraph did.

While, Sadeh (1994) initially suggested this could be a result of parental fatigue or poor parental report, he later posed another likely explanation, namely parents are only reporting the wakings that are accompanied by signalling (i.e., crying) and that the infant is in fact waking (as indicated in the actigraph readings), but that they are not crying out (Sadeh, 1996). This means that despite actigraphs limitation in detecting quiet wakings, Sadeh (1994) found that this measure was still more accurate at detecting total wake period (quiet and audible) than parental report. Further evidence for the limitation of the actigraph in detecting periods of quiet awake are discussed below.

Lichstein, Stone, Donaldson, Nau, Soeffing, Murray and Aguillard (2006) documented that in a sample of adult insomnia patients, quiet wakefulness was often mistaken for sleep. This finding has been mirrored in a study investigating a sample of pre-schoolers, where researchers reported that because the actigraph was unable to detect noise, periods of ‘awake’ were falsely labelled as sleep by the actigraph when compared to another form of measurement called videosomnography (VSG) (Sitnick et al.,2008). For example, a young boy woke and left his bed without detection from the actigraph as the frequency of movements was too low to be characterised as ‘awake’. Experimenters relied on VSG (video recordings) to confirm that the child was in fact awake as it provided both visual and auditory evidence of the night waking. It has been found that, when actigraph recordings are compared to VSG, actigraphy is more accurate in detecting periods of sleep than periods of wake (Sitnick et al., 2008).

The above findings emphasise the need to investigate behavioural interventions more thoroughly so that such limitations can be addressed. One way to confirm this is to use VSG. Videosomnography (VSG) applied to the present research involves videoing the infant from the time they are put in their basinet or cot to the time they wake and are up for the day. Depending on the technology used, videos may be time compressed (so later review takes less time) and made with infra-red illumination so as not to disrupt the normal sleep environment. Recordings are later viewed and coded to determine different sleep and wake states and to record infant and parent behaviour; including vocalisations. Anders (1979), states that this is advantageous as it allows retrospective observation of sleep.

Coding and analysis of the video record is implemented using a system that is comparable with polygraphic scoring (Anders, 1979). Using time-lapse VSG also allows researchers and families to film sleep in the natural setting of the child's home (Stinick et al., 2008). VSG data has already provided important information about the development of night waking in infants (Gaylor, Goodlin-Jones & Anders, 2000). Because VSG is able to detect wakings that are both coupled with signalling and those that are quiet and because it captures the nature and quantity of parental interactions, it provides a practical technique for assessing whether behavioural sleep interventions decrease wakings and hence change sleep-state organisation, or whether they promote self-soothing skills upon night wakings (measures as increasing periods of quiet awake).

Summary

To date, no research has reported investigating the effects of any behavioural sleep intervention on the sleep-state organisation of infants aged 6 to 12 months. While no one method is without limitations, VSG appears to provide a method that can detect if an infant is awake (both audible and quiet) or in a state of Quiet or Active Sleep, and it can detect and quantify parental interactions. VSG, therefore, used in combination with sleep diaries, makes it possible to investigate whether sleep-state organisation changes during the sleep intervention and/or whether the infant is learning how to self-soothe and hence not signalling to the parent that they are awake. While best practice sleep interventions such as the parental presence programme are known to be effective, the impact of the interventions on sleep-state organisation has been an area of research which is lacking.

Aims of the current research

The current research aims, therefore, to assess whether sleep-state organisation changes after a successful behavioural intervention for infants with ISD. The parental presence procedure was used to treat infants, and parents kept daily sleep diaries, while VSG was used to capture information on sleep-state organisation. Data analysis specifically focused on those variables that would determine, 1) whether the programme was carried out correctly and was effective and, 2) whether sleep-state organisation changed in the infants as they responded to treatment. The first was measured by establishing whether parents adhered to the programme by decreasing night-time interactions in response to infant's sleep behaviours, and whether infants responded to treatment by demonstrating a decrease in SOD (where relevant), the number of night wakings, and the duration of audible wakings. The second objective was addressed by examining whether infants increased time spent quietly awake and/or increased total time spent asleep. Furthermore, the research addressed whether these two variables apply at different phases in the infant's response to intervention. To further address the second research question it is important to establish whether sleep consolidates, resulting in fewer Active to Quiet Sleep transitions, whether Sleep Efficiency increases, and whether constructs such as LSP increase.

Method

Participants

Participants were typically developing, healthy infants aged between 6 to 12 months of age who presented with sleep difficulties including Sleep Onset Delay (SOD) and night waking. Participating families were referred by a local health services centre (see Appendix A for information sheet sent to the health centre).

After making contact, information letters explaining the study were emailed or posted to the six families who expressed an interest to their health professional (see Appendix A). Following this, each family was then contacted to further explain the study and gain more detailed information about their infant's sleep. After parents had read through the information sheet, and if the researcher established initial eligibility, they were again contacted and invited to attend an assessment interview whereby final eligibility was determined (for a list of questions see Appendix B).This included ensuring that the intervention was appropriate and safe and importantly ensured that the sleep problem was on-going and if precipitated or exacerbated by the Canterbury earthquakes that the infant was not fearful. The effect of the earthquakes on infants and their families was in part determined by their familial situation and disruption of services, including but not limited to relocation. Any appropriate disruptions are outlined later in Table 1a, b and c. In general, all parents and professionals involved felt the intervention would be of value and whilst being offered a later date all parents chose to start immediately. These parents were also sent sleep diaries (see Appendix C). Families were asked to begin filling in the sleep diaries and told that it would assist with later discussions of their infant's sleep. Three of six families continued to express an interest and were asked to come into the Pukemanu/Dovedale Centre (Child and Family Psychology Clinic) at the University of Canterbury, to attend a full assessment interview. Of the three families who did not attend an assessment interview one family stated that they were not comfortable with their infant being videoed, another family reported that their infant had begun to sleep through the night without intervention, and contact was not able to be established with the other family. Help outside of the sleep study was offered to the first two families, but not taken up.

The three participants recruited were aged 9 months, 1 week; 10 months, 2 weeks; and 7 months at the assessment interview. Information about each infant is summarised in Tables 1a, 1b, and 1c below.

Table 1a

Participant Information (Matthew)

Name	Matthew
Age	9 Months, 1 week (at assessment interview)
Gender	Male
Family Composition	Matthew lived with his mother and father.
Sleep History	He slept from 7pm until 6am in his crib for the first eight weeks. From 13 weeks his sleep became increasingly disrupted.
Night-Time Sleep	Regular routine between 6pm and 8pm (dinner, bath/massage, pyjamas, breastfeed). Matthew was placed in his cot asleep, and when he woke was reported to take 45 minutes to settle back to sleep with parental intervention. Up to seven wakings per night were reported; Matthew's mother breastfed him upon awakening and placed him in her bed for the remainder of the night (co-sleeping).
Sleeping Difficulties	SOD, night waking, night-time feeding, and co-sleeping.
Naps	Two day-time naps (20 to 40 minutes when in his cot or longer in his mother's arms).
Interventions Tried	Sleep sack, patting on back and repeating 'shh-shh-shh' until settled, breastfeeding to settle (both scheduled feeding and feeding upon night wakings), leaving to cry (both intermittently and withdrawal of all parental interaction upon night wakings).
Life Events	The recent Christchurch earthquakes had impacted the family causing stress, loss of their home, and a recent move to a nearby town outside of Christchurch. The family were located within this township for the length of the study. They were settled at the time of assessment and intervention.
Development	Typical development, described as an "active child".
Relevant Health	Minor infection at five months, ongoing topical treatment sometimes needed at night.

Table 1b

Participant Information (James)

Name	James
Age	10 months, 2 weeks (at assessment interview)
Gender	Male
Family Composition	James lived with his mother and father.
Sleep History	James had never slept through the night.
Night-Time Sleep	Regular routine between 5.00pm and 7.30pm (dinner, bath, pyjamas, breastfeed). Was placed to bed awake in cot and took 5 to 20 minutes to settle (parental intervention was sometimes needed). James was reported to wake up to six times per night. Mother attended to night wakings but he was reported difficult to soothe, until he was fed again at 5am. James could sleep up until 9am if in his parents' bed (co-sleeping) after his 5am feed.
Sleeping Difficulties	SOD, night waking, night feeding, and at times co-sleeping.
Naps	James typically had two naps that lasted 30 minutes each. He was breastfed both before and after a nap.
Interventions Tried	Tried leaving him to cry, a pacifier, and waking him for a night-time feed.
Life Events	Several family events around and after his birth (residential re-location, employment difficulties, earthquakes, death of maternal grandmother). Mother reports 'mild post-natal depression'. Living and employment arrangements were settled at the time of the study.
Development	Small as a newborn with some feeding difficulties, now developing typically.
Relevant Health	Rota virus at age 9 months, no on-going effect/s.

Table 1c

Participant Information (Flynn)

Name	Flynn
Age	7 months (at assessment interview)
Gender	Male
Family Composition	Flynn lived with his mum, his dad, and his sister (aged 3½).
Sleep History	Flynn had never slept through the night.
Night-Time Sleep	Regular routine between 5.30pm and 6.30pm consisted of (dinner, dressed in a sleeping sack, bottle of milk). Flynn either fell asleep while drinking bottled formula or was placed in his cot around 6.30pm awake. He took 20 minutes to fall asleep if happy otherwise mum rocked him to sleep. At 10.30pm, Flynn was woken for another bottle of milk. Flynn woke up to seven times per night and his parents settled him back to sleep by cuddling and rocking him. Flynn was typically up for the day at 5am.
Sleeping Difficulties	SOD, night waking, and night feeding.
Naps	Flynn had between one and two 20 minute naps a day.
Interventions Tried	Pacifier, Plunket advice: Leaving him to cry and water feeds.
Life Events	The recent Christchurch earthquakes were reported to be a stressful time for the family as Flynn's birth had to take place in Blenheim due to lack of services in Christchurch. The family home was damaged during the earthquakes and, at the time of the study it was unclear whether their house would be demolished.
Development	Typical development
Relevant Health	Reflux (treated with Losec)

Settings

Settings included the Pukemanu/Dovedale Centre (Child and Family Psychology Clinic) at the University of Canterbury for initial interviews and the family homes of the participants for all following interactions. In all but one instance the infant slept in a cot in his own room. Matthew had slept in his parents' bed since the February 2011 earthquake. During baseline, Matthew remained in his parents' bed during baseline and returned to his cot at the start of intervention.

Apparatus

During the first month a Sony DVD recorder (RDR-HX520/HX720) with an internal hard drive was linked to a video camera (with infrared capabilities) and a Pro-Mix Plus audio system (three channel stereo microphone mixer) to record the infants' sleep. The camera was attached to a suitable and safe location with adhesive strips and 3M paint tape. The camera was not placed directly over the cot to ensure safety, particularly in the case of an earthquake or aftershock. Recordings were programmed so that they began automatically without parental intervention, however, parents were asked to turn the portable audio system on when their infant went to bed and off when they woke for the day. Parents were also given eight 9V batteries over the course of contact and asked to change the batteries for the audio system every second day of its use. Regular contact was made with parents to remind them of these responsibilities. The recordings on the internal hard drive were then transferred onto Sony DVDs (240mins).

After Matthew's first intervention and James' baseline recordings were collected, the above equipment failed and new equipment was purchased. A Swann security system was used for the remaining recordings. This system consisted of two Swann Pro-Series cameras (with infrared capabilities), a Swann Advanced-Series digital video recorder with a built in hard drive (DVR4-1200), and a portable 7 Inch LCD screen. The same Pro-Mix Plus audio system was linked to this equipment. Recordings were transferred onto an external hard drive and viewed using the Swann security playback software at three times the speed of real time.

Measures

Measures used were derived from VSG, sleep diaries and Treatment Evaluation Questionnaires.

VSG Data

Sleep data from VSG was obtained via viewing on the researcher's laptop at three times the speed of real time. The coding system employed was adapted from that developed by Anders (1979). Observers were trained by their supervisor who was familiar with and trained in the coding method of Anders. Training continued until observers were competent in the coding systems. Both observers coded during 21% of the time, whilst all remaining recordings were coded by the researcher alone. The initial sleep-state was determined by observing density of movements across the first three minutes. Thereafter, transitions were detected by changes in the density of movements lasting for five minutes. In addition signalling and periods with eyes awake and focussed were recorded, as were parental interventions at the cot. Total minutes spent in each state, and the times at which they occurred (i.e., before waking) were also noted. During every minute of sleep, a state was assigned. A brief description of the coding system and corresponding sleep states is summarised below in Table 2.

Table 2

Classification of Sleep and Wake States for Coding Infants Night Sleep Behaviours

Code	State and Description
0	Sleep that is not otherwise specified.
1	**Quiet Sleep** is characterised by an absence of body movement (Anders, 1979; Thoman, 1989; Anders & Keener, 1985; Halpern, 1995). Onset of Quiet Sleep is determined by an absence of body movement for three minutes. Quiet Sleep continues until the infant transitions into another sleep or waking state. Occasional startles are observed during this sleep state.
2	**Active Sleep** is characterised by sudden, involuntary body movements (see Anders, 1979; Thoman, 1989; Anders & Keener, 1985; Halpern, 1995). The infant must make two movements within three minutes for Active Sleep onset, followed by two movements every five minutes to classify continuing Active Sleep.
3	**Awake/Talking** is characterised by any length of time that the infant is awake and talking.
4	**Awake/Quiet** is characterised by any length of time that the infant is awake and quiet (i.e., eyes open and alert). This includes self-soothing behaviours (i.e., thumb sucking).
5	**Crying/Awake** is characterised by any length of time that the infant is awake and crying.
6	**Parental Interaction** is characterised by any length of time that the infant is engaged with parent or out of the cot.

NB: Duration of time spent in each sleep or wake state is recorded in whole minutes

In addition to the variables stated above the researcher also calculated the following variables: SOD (minutes), Number of Night Wakings, Number of Sleep-State Transitions (Active/Quiet), Number of Sleep-Wake Transitions, Sleep Efficiency, Longest Sleep Period (LSP), and Largest number of Active-Quiet Sleep-State Transitions before Waking. The rationale for including this last measure is that like LSP it provides a measure of uninterrupted sleep and could suggest changes in sleep consolidation. Unlike LSP, this measure illustrates the largest number of sleep-state transitions (Active-Quiet Sleep) an infant sleeps through before waking. This measure was considered appropriate after analysing the results.

Parent Report Data

Parents were given sleep diaries adapted from France (1989) (see Appendix C). The diaries contained spaces for parents to record their infants sleep using the following constructs: time, location and duration of day-time sleep; evening bed-time; the duration of any initial settling time (SOD) and the nature of any associated crying or distress; the number and duration of any night waking; and parent actions in response to any night waking (France, 1989). The sleep diaries were completed by at least one parent in each family daily until the cessation of each follow-up.

Furthermore, to assess clinical application of the intervention, an Infant Sleep Disturbance (ISD) score was calculated for each phase of the study. This was calculated using an adaption of Lawton and colleagues (1991, utilised by Healey, France, & Blampied, 2009). The ISD scores were calculated using sleep diary data over the last four nights of each phase. Infants were scored a 1 (yes) or 0 (no) on three criteria: whether SOD exceeded five minutes, if infants woke more than once, and if the total wake time exceeded ten minutes. A sleep disturbed infant would score highly (12/12) while an infant without ISD would score low (0/12).

Parents were also given a Treatment Evaluation Questionnaire to complete (see Appendix D). This is a nine-item questionnaire adapted from Kazdin, 1980 (as cited in, Kelly, Heffer, Gresham & Elliot, 1989) and was administered to parents after the follow-up phase of the study. The purpose of the questionnaire was to measure parents' satisfaction with the effectiveness and accessibility of the interventions implemented (Selim et al., 2006). A score of 31 to 45 represents strong satisfaction and low stress, a score of 16 to 30 represents moderate satisfaction and stress, while a score of 1 to 15 represents low satisfaction and high stress.

Design

A multiple-baseline-across-participants design was employed (Cooper, Heron & Heward, 1987). All participants received the same independent variable (the parental presence procedure). Each family moved through the following phases: Phase 1: Baseline of a minimum of three days and increasing to seven to ten days across successive participants. Phase 2: Parental presence sleep programme for 28 nights (see description under Procedure, below). Phase 3: A maintenance phase for approximately a month. During this phase parents responded to each of their infant's night wakings in a low key fashion, intervening only if they considered the infant had a need for parental attention. This was followed by Phase 4: Follow-up for seven nights. At this point the sleep diaries were collected and the Treatment Evaluation Questionnaire was administered. Sleep diaries were collected for each night of the study (approximately two and a half months). Video recordings were programmed for two to five nights during baseline, eight nights across two occasions during intervention and two further nights during the follow-up phase.

Procedure

Ethics

Prior to beginning the present study, a proposal was sent to the Human Ethics Committee at the University of Canterbury, New Zealand. Ethical approval was gained on the 27/09/10 and amendments were again approved on the 05/05/11 after changes were made in regards to the limitations imposed by the Canterbury earthquakes (see Human Ethics letters of approval in Appendix E). Informed consent by parents was gained for each participant before the commencement of any data collection or intervention (see Consent Forms in Appendix F).

Assessment and Induction Interview

This initial parent interview was conducted by the researcher (a trainee Child & Family Psychologist) and Senior Supervisor who is a registered Clinical Psychologist. Confidentiality limits were described and the interview took place with parental consent. The assessment interview was semi-structured with questions addressing various domains of the infants' past and current development, with considerable attention focused on the infants sleep problems and past treatments (see Appendix B). Researchers also cautiously investigated the each family's experience of the Canterbury earthquakes and established whether the intervention would be appropriate or clinically safe at the time of the study. Researchers also answered questions, informed the parents of the procedures to be followed, and familiarised the parents with the sleep diary. Formal consent for participation was gained once parents had sufficient time to decide whether they wanted to take part in the study and before any baseline data was collected (see Appendix F).

Phase 1: Baseline Data Collection

During the assessment interview parents were asked to randomly pick a card that represented the length of baseline. Baseline lengths included, ten nights (five nights visual recording); nine nights (four nights visual recording); eight nights (three nights visual recording); and seven nights (two nights visual recording). Matthew was randomly assigned to a baseline length of nine nights, James seven nights, and Flynn eight nights. During the length of baseline parents were asked to record sleep behaviours each night using the sleep diary provided.

Phase 2: Intervention Training Procedure (Parental Presence Programme)

After baseline data had been collected, a subsequent meeting took place whereby the sleep programme was discussed. This was a collaborative discussion between the researcher and parents. The discussion covered any anticipated problems, for example, parental concerns about implementing the sleep programme, and possible solutions were discussed. At the end of this interview a schedule for daily then weekly telephone calls was negotiated and brief instructions on the sleep programme were provided.

The parental presence procedure, as selected for each infant after this discussion, was administered by the parents for their child for a minimum of 28 days. Parents were instructed to do the following at bed-time (adapted from Selim et al., 2006):

> “When you put you baby to bed at night, put him or her in the cot, kiss goodnight and then lie in the alternate bed in that room pretending to be asleep. Remain like this until your child has settled to sleep. At this point you can quietly leave the room and resume your evening activities. If your baby wakes and cries, return to the room. Do not interact with your baby but go straight to the alternate bed and pretend to be asleep, until your child has settled. Throughout this period do not attend to your baby directly unless he or she is ill or in danger. If the baby is in danger (for example during an earthquake or aftershock, or when he has a leg in the bars of the cot) respond to this appropriately by decreasing hazards and ensuring safety. If your child is ill or there is a significant earthquake you will need to halt the programme and resume when your child is well again or things are settled. Remain sleeping in your baby’s room for seven nights then return to your own room. Usually the baby accepts this change readily. Occasionally the baby will resume crying when you return to your room. In this case return to the baby’s room for a night or two, this usually corrects the problem. When you have returned to your room, carry on the programme for a further

three weeks. Throughout the programme, treat all crying prior to 6am as a night waking and all crying after 6am as 'up' for the day. Once baby is up for the day increase light, sound and activity to enable your child to discriminate night and day."

Phase 3 and 4: Maintenance and Follow-Up

Approximately five weeks after the behavioural intervention, and once parents were satisfied with progress, any video equipment remaining in parents' houses was collected. During this final meeting parents were given a maintenance programme instructing them to check their infants upon night wakings in a minimally intrusive way, attending to their infant if there was a good reason, such as a fright or wet nappy, and otherwise bidding "goodnight" and leaving the room. A final follow-up then took place approximately one month from this date. In follow-up, the sleep diary was completed for one further week and two further nights were recorded on video. At this time a Treatment Evaluation Questionnaire was completed.

Results

This section is structured as follows. First, factors affecting the intervention and quality of the data are discussed; Second, the extent to which the infants' sleep responded to the intervention is assessed, primarily using data from the sleep diaries, although VSG data is used to measure duration of parent interventions as a measure of compliance with the programme. Third, changes in sleep-state organisation are assessed using data obtained from video recordings. Finally, satisfaction of the parents with the programme is reported.

Factors Affecting the Intervention

All families completed a parental presence programme. Programme and phase lengths achieved varied depending on external factors such as sickness, travel, parental adherence, and earthquakes/aftershocks. Due to the earthquakes around Canterbury at the time of the study some hours of visual recording were lost across all participants owing to power cuts. Despite many aftershocks, none of the infants were reported by their parents to wake with a fright throughout the course of the present study. Nights on which there was disruption due to the child's illness, major disruptions to the family, and/or non-compliance with the programme are shown on the figures (indicated by *). These factors were considered as significant enough to impact on execution of the intervention (i.e., noncompliance) or its appropriateness (i.e. sickness), and where appropriate descriptions of nights of disruption are further explained. The impact of these factors on each infant is outlined below.

Matthew

Half a night of visual recording was lost for Matthew due to equipment failure during baseline phase (night 12). During the intervention equipment failure (night 28), parental non-adherence, and travel (away from the family home) meant that the intervention was unable to follow the structured format proposed and instead an extended programme was implemented.

Specifically Matthew's mother began sleeping in his room on night 18, and once Matthew slept through the night without audible night waking she returned to her own room. Matthew had consecutive nights of disrupted sleep on nights 46 to 49, his mother was advised to return to the initial stages of intervention and slept in his room from night 50 to 60 of the study, until his sleep became more settled. Parental non-adherence included picking Matthew up, cuddling, and breastfeeding him. These behaviours occurred approximately once a night every seven days. This was typically in response to a spontaneous increase in Matthew's sleep disturbance.

James

There were unanticipated problems gaining access to the video equipment during the baseline for James, and so there were only two video recordings made in this phase. During the first week of intervention, James became ill on night 11 and the programme was suspended until night 13. In the early part of the resumption of the programme James continued to recover from his cold. James' mother also reported that the number of night wakings during baseline was unusually low and not what she would have expected. Furthermore, partial data (less than two hours) was lost on the following nights 22, 23 and 36 due to equipment failure.

Flynn

Baseline progressed as anticipated with the exception of one hour of missing video data due to equipment failure (night eight). During the first week of intervention, Flynn's parents reported a dramatic decrease in sleep disturbance and in fact reported minimal night wakings and little difficulty with sleep onset. However, after three nights of intervention, Flynn began sleeping similarly to baseline and thus his parents were advised to treat night four as the first night of intervention and to continue to sleep in his room for a further seven nights. The programme then continued as proposed, however further equipment failures were noted on nights, 19, 32 and 33. Unfortunately during the follow-up phase, Christchurch experienced a large earthquake which was reported to minimally disrupt Flynn's sleep thereafter and meant that only one night of video data was obtained during the follow-up period. Due to significant stress the family chose to sleep elsewhere and this meant that sleep diary recordings were also limited.

Quality of the Data

All parents were supplied with sleep diaries that they could complete prior to their assessment interview. While Flynn and James' parents did not complete the full diary (only included minimal and insufficient data), Matthew's parents completed several extra days and therefore baseline sleep diary data was obtained for a longer time than planned. Intervention lengths for all three participants changed according to sickness or unavoidable circumstances (such as equipment failure). Sleep diary data was obtained throughout baseline, intervention, maintenance and follow-up. There is missing sleep diary data for all three participants within the follow-up period.

Video recording data (or VSG) was obtained for a portion of each phase for all three infants (see Table 3 below) by way of recordings made each night for up to four consecutive nights. Each infant's sleep was visually recorded for a portion of consecutive nights across baseline. During the intervention phase, for each child videos were obtained for four consecutive nights in the second and fourth week. A proportion (one to two nights) of follow-up was also videoed approximately one month after the last week of intervention.

Table 3

Proportion of Video Recordings

	Baseline	Intervention	Follow-Up
James	2/8	8/31	2/7
Flynn	3/10	8/37	1/7
Matthew	4/17	7 ½/45	2/7

Comparison of Parent Sleep Diary and VSG Data

Infants' response to intervention was measured first by changes in SOD, number of night wakings, and duration of waking as recorded by sleep diary data and second from VSG data. To check agreement between parent diary data and VSG, those variables measured by both VSG and parental report were compared for every night where both measurements were used. Nights were coded as 'agree' or 'disagree'. For SOD and duration of crying, reports were considered 'agree' if the difference was less than ten minutes. Number of audible night wakings was considered 'agree' if the difference between the two reports was two or less. The number of agreements was then divided by the number of agreements plus disagreements. This figure was multiplied by 100 to give the figures outlined below in Table 4. Flynn's parents estimated duration of awake and crying retrospectively (weeks after) and hence this data was not included.

Table 4

Reliability of Sleep Diary Data Compared to VSG

	Matthew	James	Flynn
SOD	67%	100%	100%
Audible Night Wakings	79%	78%	73%
Duration of Waking	56%	89%	N/A

NB: The parents' measurement of SOD varied dependent on the phase of the study. For example during baseline for Matthew his mother slept with him and therefore could measure SOD from what she saw and heard. Flynn and James' parents were out of sight from their infants and therefore measured SOD as audible crying. Furthermore at the beginning of intervention, parents remained in the room with their infants and measured SOD as the state change to sleep.

When compared this way, VSG and sleep diary data are moderately to very congruent for SOD, number of night wakings, and duration of waking. Discrepancies between the two measurements are likely due to what either was not filmed by the camera (SOD during baseline) or not observed by the parent (i.e., quiet wakefulness). For instance, SOD and number of night wakings are influenced by periods of quiet wakefulness which are typically only recorded in the VSG data (except as explained in the footnote to Table 4). The percentage of agreement for each variable does however suggest that sleep diary data alone provides an accurate measurement of those sleep variables that can be heard or observed by the parents.

Parent Interaction

Parent interaction with their infant, as measured by VSG (see Figure 1), provides a measure of compliance with the intervention protocol (on the nights recordings were made), and ideally should be of short or zero duration as of the beginning of intervention. In baseline, all parents spent considerable time each night interacting with their infant. Interactions observed during baseline included, feeding, cuddling, rocking, and changing infants. Across intervention and follow-up, parental interaction was minimal with most nights illustrating no parental interaction for all three infants. In summary, this data illustrates that for Flynn and Matthew, parental interaction at night was lengthy before intervention. Across intervention, parental interaction dramatically decreased and changes remained consistent at follow-up.

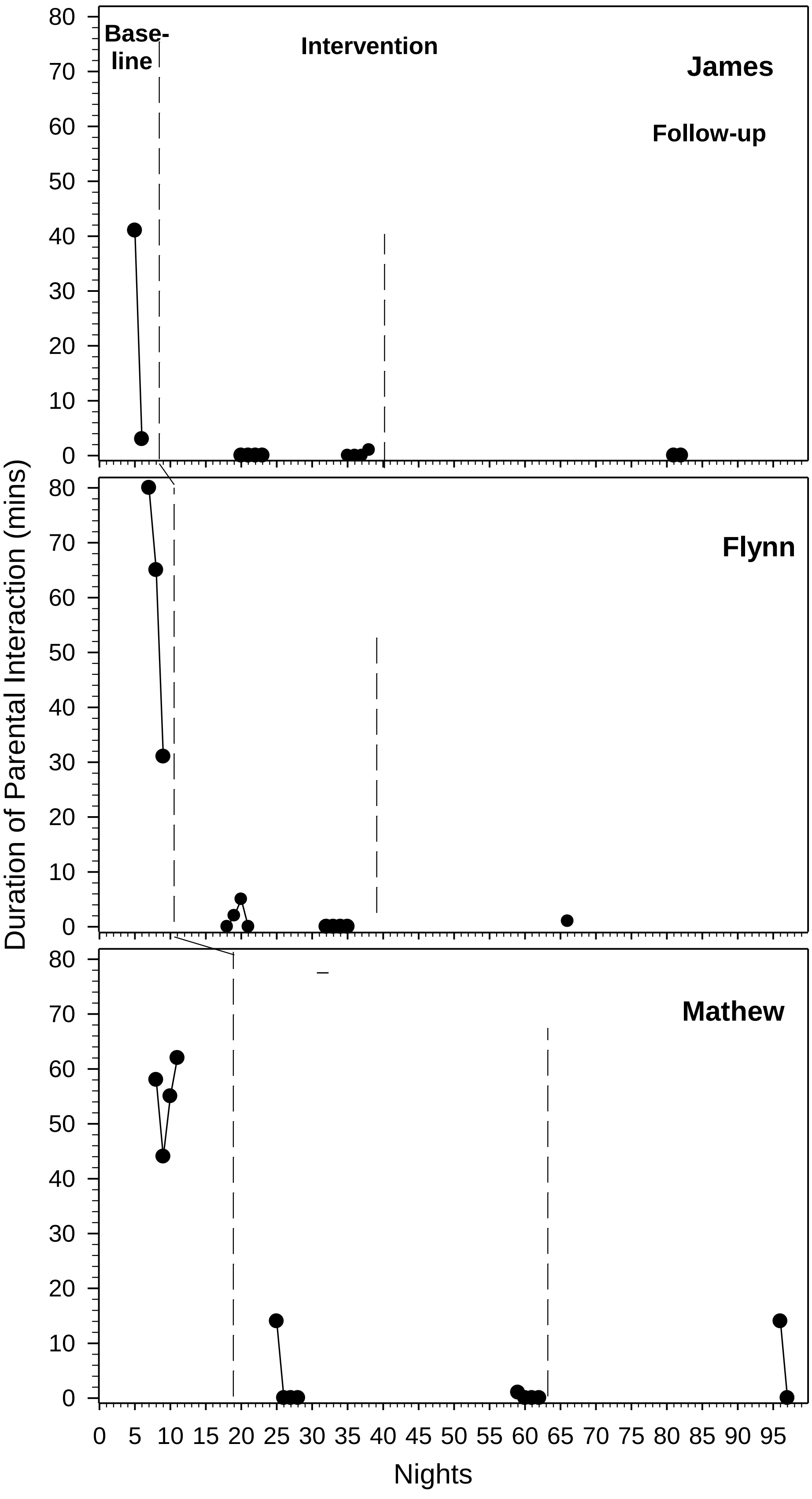

Figure 1. VSG data of duration of parental interaction

Infant Response to Intervention

Treatment effectiveness (assessed by sleep diary data) would be indicated by changes in one or more of the following: a decrease in SOD, a reduction in night wakings, and a reduction in the duration of audible night wakings. Sleep diary data is presented below to assess whether each infant's sleep improved after intervention.

Sleep Onset Delay (SOD)

All three infants were reported to have difficulties with sleep onset, either not being able to fall asleep without parental input (James, Flynn & Matthew) – in which circumstance observed SOD may be shorter than it would be if it was assessed as independent sleep onset - and/or with lengthy SOD (Flynn & Matthew) if sleep initiation was attempted without active parent management. SOD is illustrated in Figure 2. Typically James' SOD was short (0 to 6 minutes) across baseline, intervention, maintenance, and follow-up. During this phase, James' mother put him to bed asleep for four of the nights which accounts for the nights of no delay. James' longest SOD (86 minutes) was during the first night of intervention. With the exception of one disruptive night and a night of illness, James' SOD was generally short and stable.

Flynn's SOD data shows a clear intervention effect for although the baseline data show some reductions in SOD over the last four nights, during the first half of intervention, Flynn's SOD waxes and wanes between periods of delay (10 to 16 minutes) and little or no delay. During the second half of intervention Flynn's SOD decreased to less than a minute and then remained relatively stable over the maintenance phase. Periods of short SOD (less than four minutes) increased in frequency as intervention progressed with very few nights of disruption. On one of the disruptive nights Flynn was sick (night 52). Furthermore, most nights of disturbed sleep were less than that observed during baseline. During follow-up

Flynn’s SOD increased, as Christchurch experienced a number of small aftershocks during this period.

Matthew’s SOD during baseline varied between periods of short and long delay. This pattern continued initially in the treatment phase, but stabilised somewhat by the second week of intervention. After this point, Matthew experienced an increase in the frequency of short SODs that were notably shorter in comparison to his baseline levels. With the exception of a small number of long SODs, Matthew’s SOD typically fell under six minutes in length. It is important to note that each period of increased SODs became spaced out over time and the magnitude of SOD tended to decrease over these episodes. In some cases the deterioration in SOD can be related to events. For example, on night 37 Matthew’s mother began responding similarly to how she had during baseline (i.e., cuddling and breastfeeding). On night 47 and 48, the family went away on vacation and Matthew’s sleep became disrupted as a result so his parents were advised to revert back to intervention from night 50 until night 56. On nights 80 and 87, Matthew again returned from holiday, and on night 94 he became ill.

In summary Flynn and Matthew showed a treatment effect with a notable reduction in SOD during maintenance. James’ SOD was not problematic in that he did not take long to fall asleep (or was placed in bed asleep) before intervention and this remained consistently low throughout the course of the study.

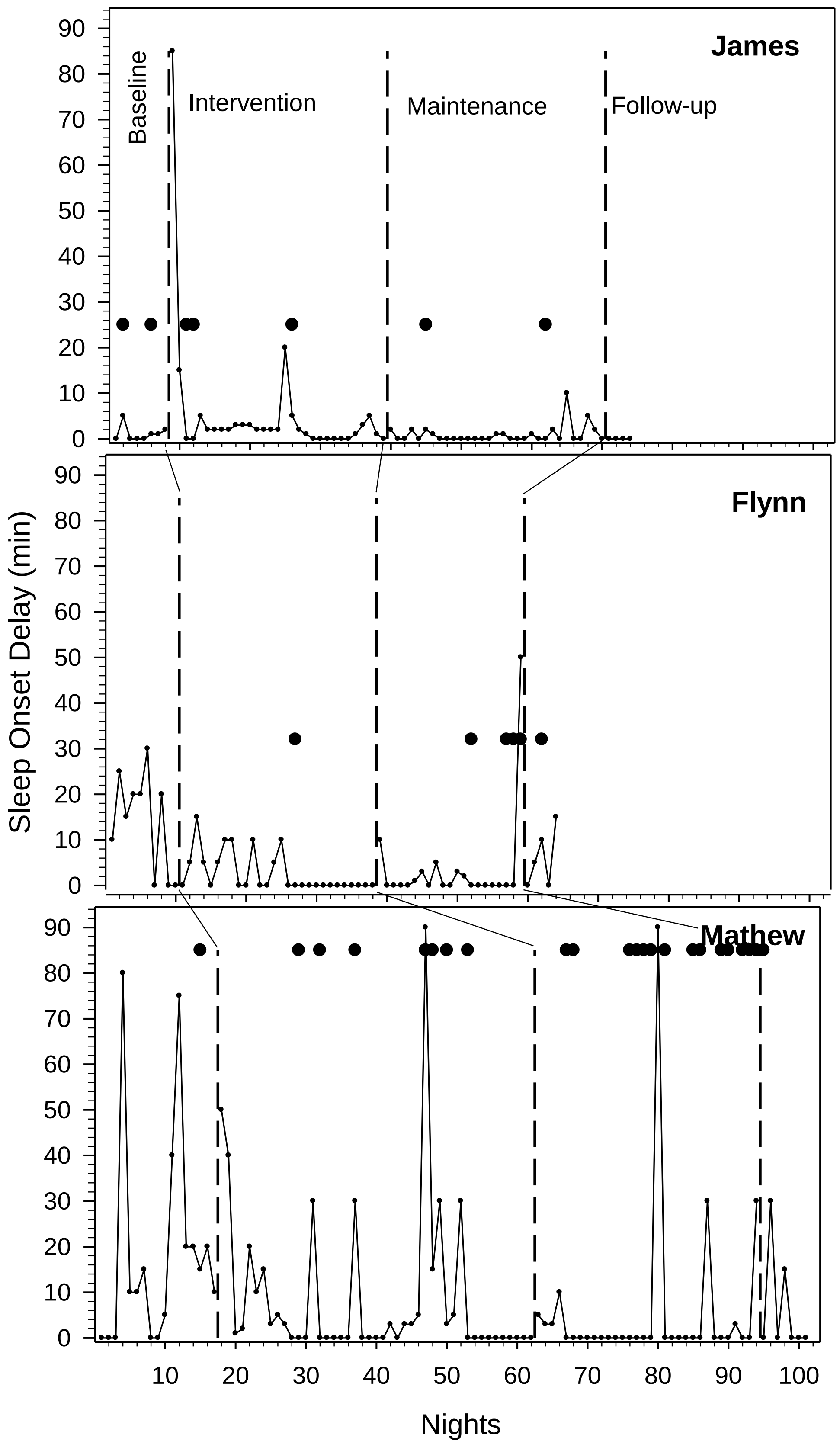

Figure 2. Parental report of Sleep Onset Delay (SOD)

Number of Wakings per Night

Number of wakings per night are shown in Figure 3 below. During baseline James woke between 1 to 12 times per night. During the first intervention night he woke eight times a night, but waking trended down thereafter. Increases in waking on some nights are clearly related to illness or other disruption in routine (Figure 3). Over the intervention phase, nights when James woke less than twice increased in frequency as intervention continued. The observed reduction in night waking continued during maintenance with James typically waking up to two times a night with exceptions on nights of sickness or disruption. During follow-up James woke once or twice per night indicating that his decrease in night wakings remained stable.

During baseline Flynn woke between two and seven times per night, but mostly between two and four times. During the first two nights of intervention, when his father slept in his room, Flynn did not wake, but this effect was short-lived. This pattern continued in maintenance and at follow-up, despite several nights of illness or other disruption (Figure 3).

During baseline Matthew woke between five and eight times per night, with an increase in waking frequency associated with illness (Figure 3). Night wakings reduced during intervention and Matthew was noted to first sleep through on night 18 of intervention. Thereafter he typically woke once per night, with some increases in waking frequency associated with illness or disturbance (this family experienced considerable disruption; see above). During maintenance, if Matthew woke it was typically once a night with this reduction in waking being maintained despite some nights of illness or disruption, and this remained the case at follow-up.

In summary all three participants woke often during baseline indicating ISD expressed as night waking. A reduction in wakings per night was replicated for all three participants after intervention and remained stable at follow-up. Notably, for Flynn and Mathew, the improvement in waking was observed despite numbers of nights (a large number for Mathew) where illness or family disruption was experienced.

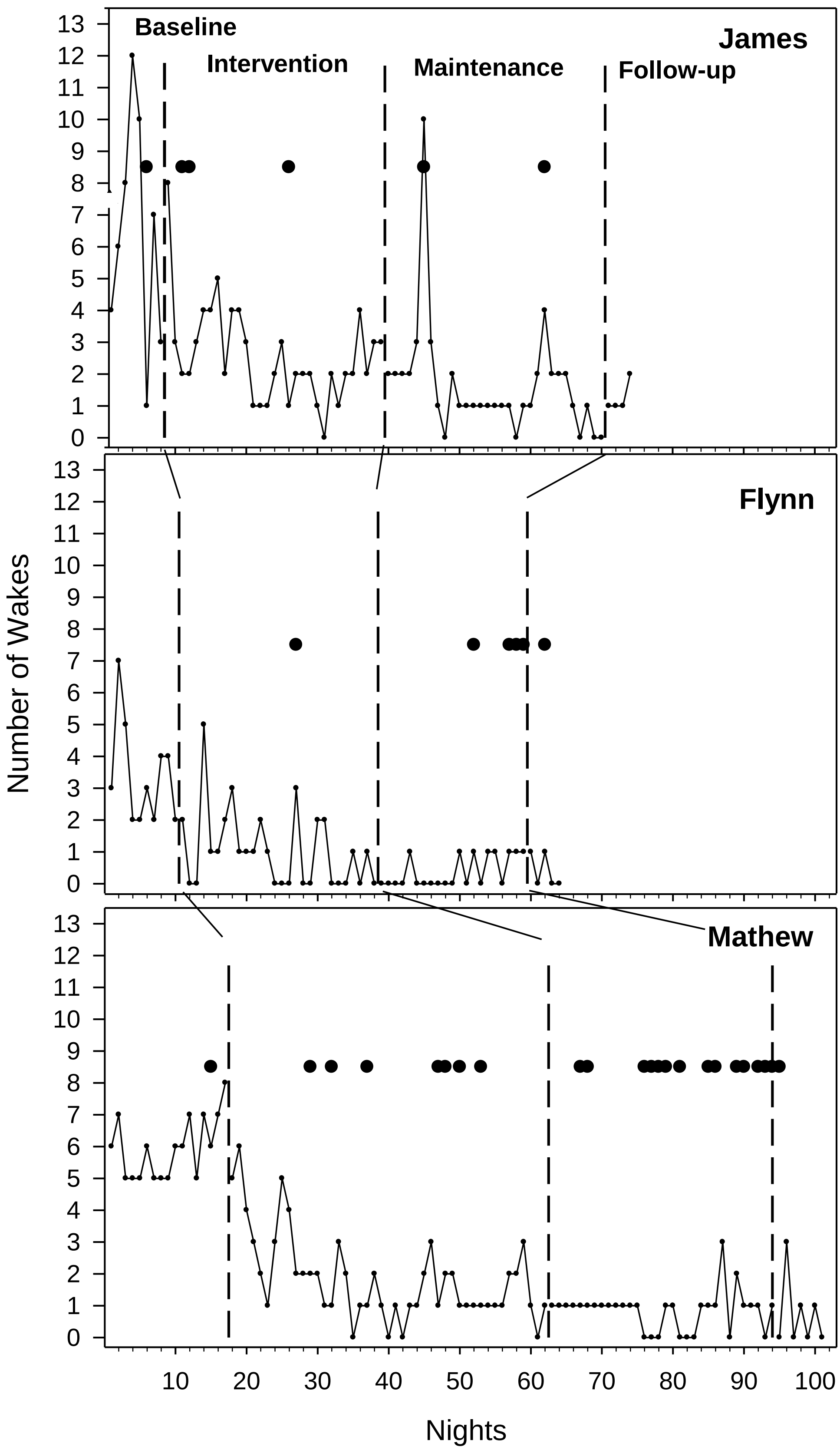

Figure 3. Parental report of number of night wakings

Duration of Audible Wakings

Figure 4 illustrates the duration of audible waking (i.e., those that attracted the parent's notice because of crying and calling out) across each phase of the study. As Flynn's parents estimated the duration of waking (as explained above), their data is not included in the figure. James' woke for increasingly long periods in the first part of baseline, associated with nights of illness or disruption, and then decreased his durations, but always exceeded more than 30 minutes. During intervention, James' duration of audible wakings steadily decreased and, importantly, nights of little or no audible waking increased as intervention progressed. This was maintained after intervention with the exception of one night of illness or disruption during the maintenance phase, and continued at stable levels lower than those in baseline at follow-up.

The duration of wakings during baseline typically fell below 90 minutes for Matthew but the trend over baseline was for wake duration to increase. During intervention, the maximum duration of time spent awake dramatically increased and became very variable. Although the frequency of his waking tended to decrease (see above), the duration of each waking episode was often long, and sometimes very long, with little evidence of improvement in this pattern over the intervention phase. During maintenance phase, improvement by way of both reductions in duration and variability of waking becomes evident, especially in the second half of the phase. With the exception of one night, this improvement is sustained at follow-up.

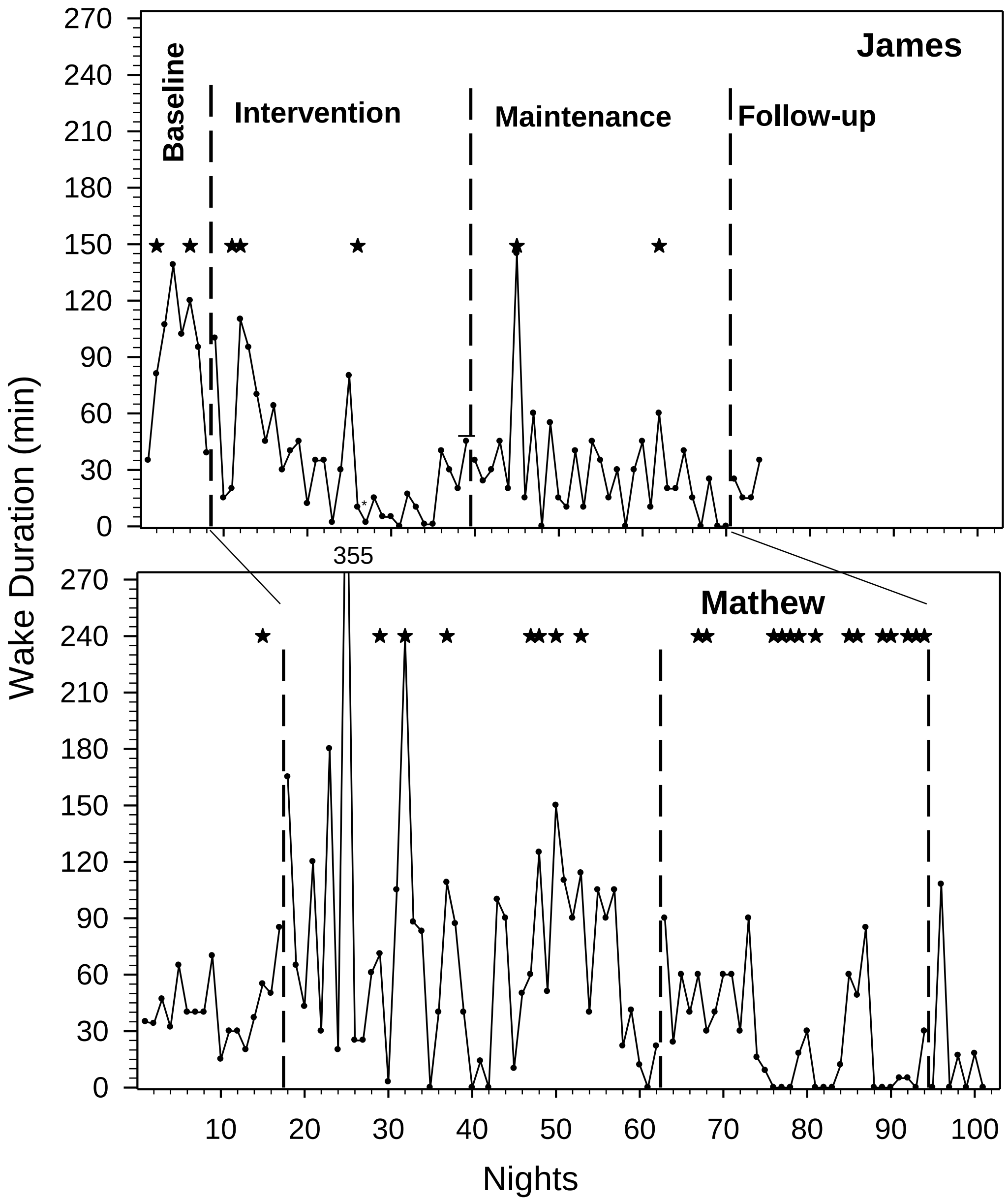

Figure 4. Parental report of duration of audible night wakings

Clinical Application

The results illustrate an improvement in sleep as a response to intervention. To assess the clinical impact a composite ISD Score was derived for each participant across baseline, intervention, and follow-up using the procedure described in the Method section above. The ISD scores for each participant are represented for each phase in Table 5. ISD scores decreased for Flynn and Matthew to a level indicating an absence of ISD by follow-up. James' scores decreased a little by follow-up. As SOD was not problematic for him (in terms of duration), his ISD score at baseline is lower compared to Flynn and Matthew.

Table 5

Clinical Impact of Intervention According to ISD Scores

	Baseline	Intervention	Follow-up
James	7/12	8/12	5/12
Flynn	9/12	1/12	2/12
Matthew	12/12	4/12	3/12

Summary: Effectiveness of Intervention

All three participants responded to treatment somewhat differently, particularly in terms of how rapid any improvement was, but favourably, illustrating that the sleep programme had improved aspects of these infants' sleep. James' SOD in terms of duration and duration of crying was not regarded as problematic previous to intervention and remained relatively stable across intervention with the exception of the first night of intervention. In general, his nightly wakings steadily decreased across intervention and remained stable during maintenance and follow-up. The frequency of nights sleeping through without waking also increased as intervention progressed.

Flynn showed a clear treatment effect. His SOD and the number of night wakings decreased as intervention progressed. Nights of sleeping through also increased with time. Duration of crying increased during the first half of intervention and decreased to minimal levels by follow-up.

Matthew showed a clear trend of nightly wakings decreasing with time. His SOD waxed and waned typically in response to some disruption in routine. Each peak however, reduced as intervention progressed. Duration of crying increased during the first half of intervention and then decreased considerably during maintenance. These changes remained stable across follow-up.

Changes in Sleep-State Organisation

Sleep-state changes were measured by VSG for each participant across baseline, intervention, and follow-up. The following variables were measured: Total Duration of Awake (Quiet and Audible), Duration of Quiet Awake, Number of Sleep-Wake Transitions, LSP, and Largest Number of Active-Quiet Sleep-State Transitions before Waking.

Inter-Rater Reliability

To measure the reliability of coding, eight out of thirty eight (~21%) videos were chosen at random and reviewed by a second rater. The second rater was asked to code each minute of sleep following the system reported in the Method section. These were then coded as ‘agree’ or ‘disagree’ against the original coding sheet. The number of agreements was then divided by the number of agreements plus disagreements. This figure was then multiplied by 100. This is referred to as the Inter-Rater Reliability Index. Reliability for each video and the mean Inter-Rater Reliability Index is shown below (see Table 6).

Table 6.

Inter-Rater Reliability

Video	**Reliability (%)**	**Participant and Phase**
1	78%	Flynn Baseline N2
2	93%	Flynn Intervention W2 N4
3	89%	Matthew Baseline N3
4	98%	Matthew Intervention W1 N2
5	88%	Matthew Intervention W4 N1
6	86%	Matthew Intervention W4 N2
7	89%	James Baseline N2
8	89%	James Follow up N1
Average %	89%	

NB: W=Week, N=Night

Duration of Awake

Changes in the Total Duration of Awake and time in Quiet Awake across each phase of the study is illustrated below in Figure 5. Total Time Awake is the sum of Quiet Awake and any other time spent waking, but not quietly. In Baseline, the pattern of change in the two time measures tends to be the same, with Total Time Awake considerably longer than Quiet Awake time, although one observation for James and one for Flynn show convergence. For James, Total Time Awake during intervention and at follow-up is quite variable, but somewhat shorter than the maximum observed in baseline. Quiet Awake time is low across these two phases, so that James appears to be continuing to spend considerable time awake, little of it quiet. Flynn's data shows a clear intervention effect as Total Time Awake decreases dramatically, in comparison to baseline, by the end of intervention. Total Time Awake and Quiet Awake are similar at the end of intervention and at follow-up suggesting that when awake, Flynn spent less crying or signalling to parents and proportionally more time quietly awake. Matthew's data show a similar response with Total Time Awake decreasing at the end of intervention. Data show convergence of Total Time Awake and Quiet Awake which is maintained at follow-up.

In summary, Total Duration of Awake per night reduced for Flynn and Matthew who both had significant difficulties with the amount of time spent awake during baseline. James does not show systematic changes in either wake time, with interpretation slightly unclear due to a limited numbers of baseline recordings. Overall, for none of the three infants, is their evidence that the absolute duration of Quiet Awake increased over the phases of the study, however there is a relationship evident for two infants (Flynn and Matthew) in which Quiet Awake increased and then decreased to levels equal or below baseline. Furthermore, for these two infants, Quiet Awake is proportionally larger during the end of the study in comparison to Total Awake, as compared to baseline.

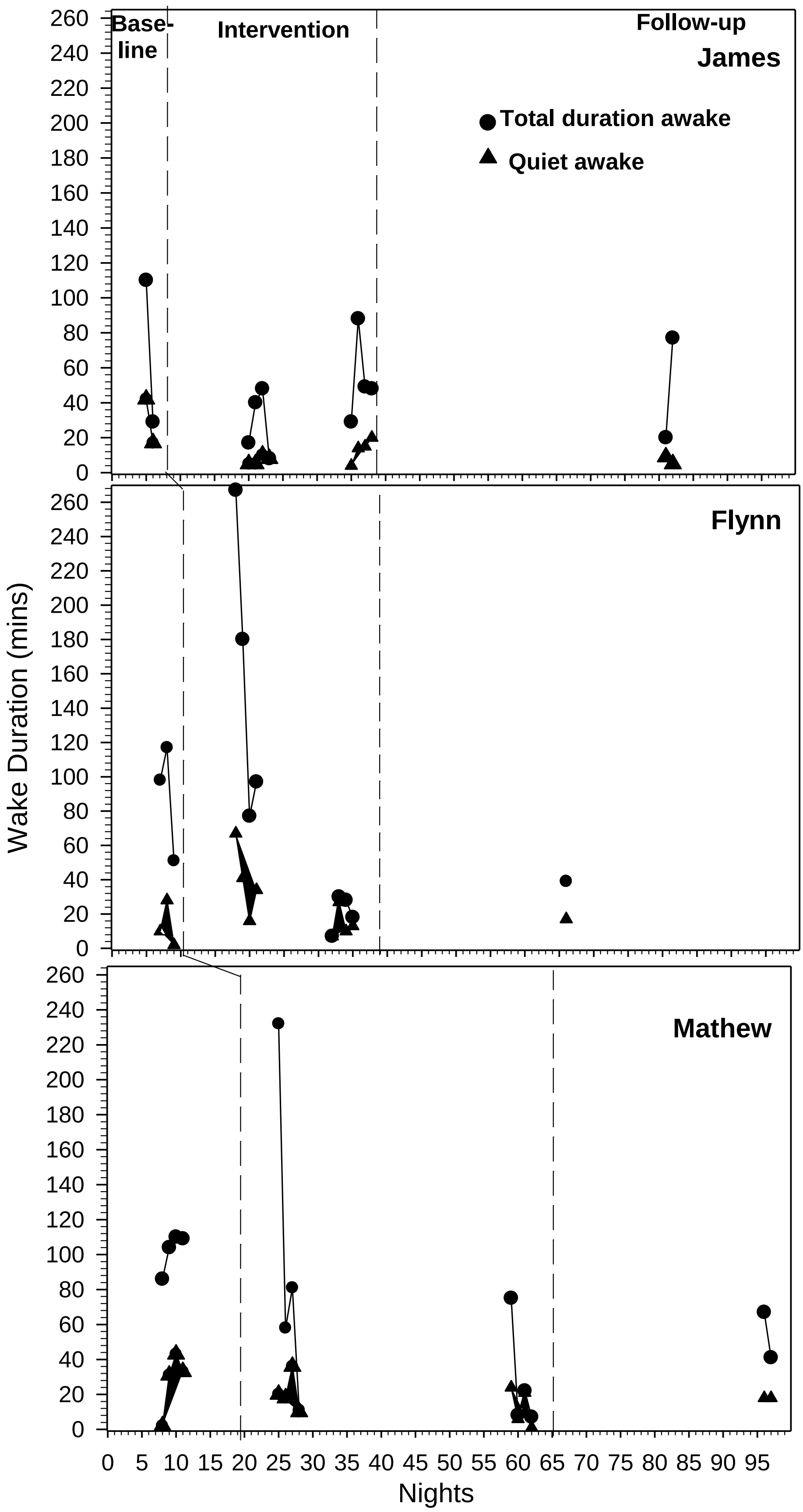

Figure 5. VSG data of duration of time spent awake and quiet compared with total time awake

Sleep Efficiency

Sleep Efficiency changes are illustrated in Figure 6. These scores were taken at the designated time points during visual recording (see Method). Importantly, each recording occurred after the same amount of time had lapsed in intervention, maintenance and follow-up. James' Sleep Efficiency scores remained relatively high across the study indicating that James spent a high percentage of the time he was in bed, asleep. This suggests that this was not a problematic area for James before intervention.

Flynn's Sleep Efficiency scores are moderately high (at 80 to 90%) during baseline. This suggests that on some nights Flynn was awake for up to 20% of the night. During the first part of the intervention his Sleep Efficiency scores dramatically decreased indicating an increase in the proportion of time spent awake while in bed, but by the end of the intervention phase, his efficiency had levels slightly above that during baseline. This suggests an increase in the proportion of time spent asleep and this increase remained stable at follow-up.

Matthew's data also replicates what was noted in Flynn's. During baseline, Matthew's scores ranged from 75 to 85% which suggests that Matthew spent a relatively significant proportion of his night in bed awake. During the start of intervention Matthew's scores decreased and then dramatically increased during the second week of intervention. The increase is slightly above what is illustrated during baseline and this remained stable at follow-up. Matthew's data does suggest an improvement in Sleep Efficiency suggesting a lower proportion of his night is spent awake and higher proportion is spent asleep.

For Flynn and Matthew, who both spent a large proportion (10 to 25%) of their night awake, their Sleep Efficiency scores increased suggesting some improvement in the proportion of time in bed spent asleep. James did not have notable difficulties within this area before intervention; and his Sleep Efficiency remained stable across all phases of the study.

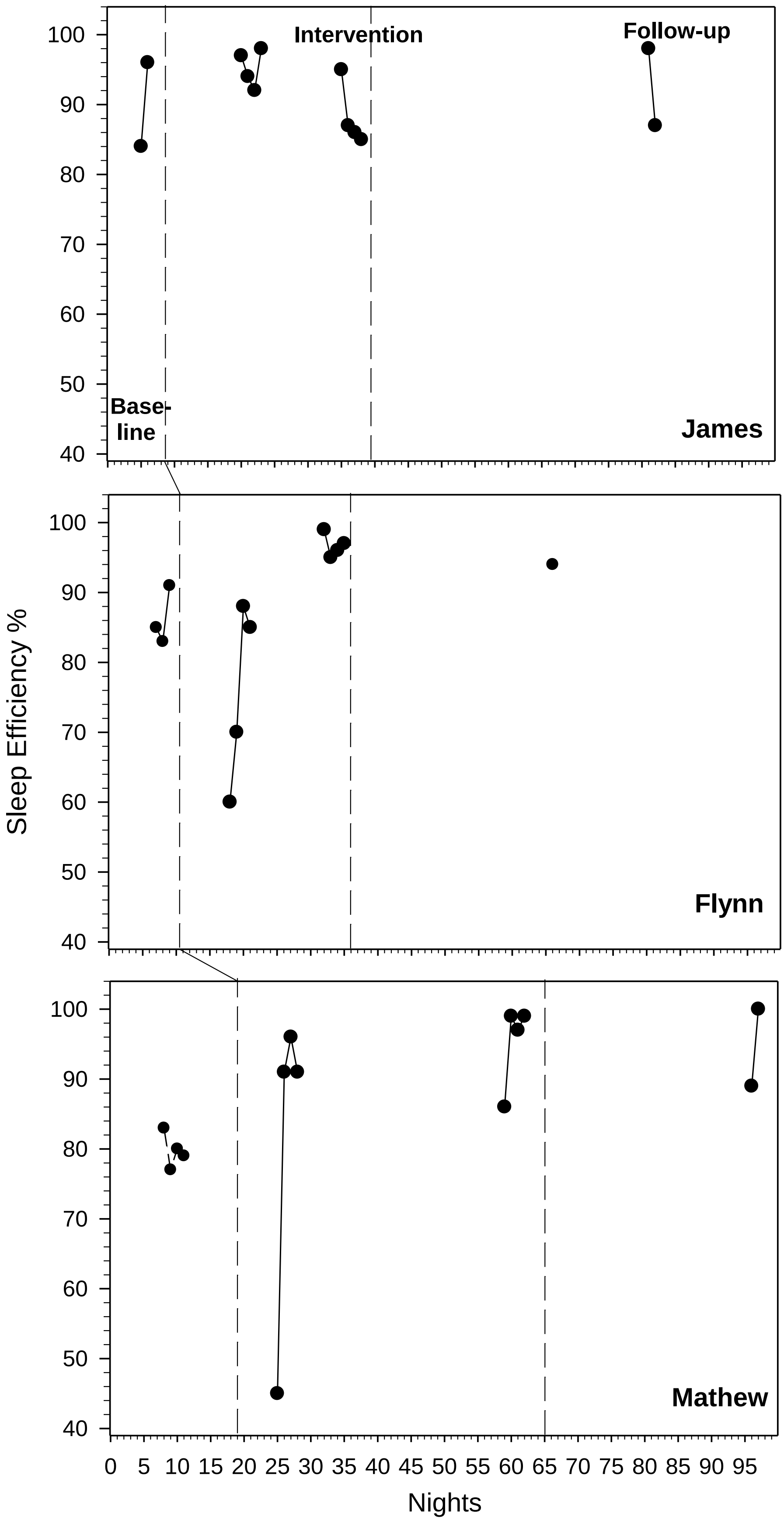

Figure 6. VSG data for Sleep Efficiency

Longest Sleep Period (LSP)

Longest Sleep Period (LSP) across baseline, intervention and follow-up is illustrated for each participant in Figure 7. For all three infants LSP during baseline is typically less than 50% of the night. James' baseline recordings illustrate that LSP before intervention ranged between 180 to 330 minutes a night. LSP increased to between 390 to 630 minutes during intervention and this change remained stable at follow-up.

Flynn's LSP at baseline deteriorated indicating a decrease in the length of sustained sleep before waking. At intervention Flynn's LSP steadily increased with time with the exception to one outlier which occurred on night 33 when two hours of visual recording was lost due to power cuts. This is likely to have impacted what could be calculated for Flynn's LSP. At follow-up this change did not continue and it should be noted that this period of time coincided with Canterbury earthquakes.

Matthew's LSP at baseline was the shortest of the three infants ranging between 90 and 150 minutes. At intervention LSP varied during the first half of the intervention phase however, by the end of intervention LSP had increased to between 250 and 450 minutes. These changes continued at follow-up.

In summary, the data presented in Figure 8 illustrate that LSP increased for all three infants at intervention and that they could sleep for a longer duration before waking in the night or for the day which suggests the development of sleep consolidation.

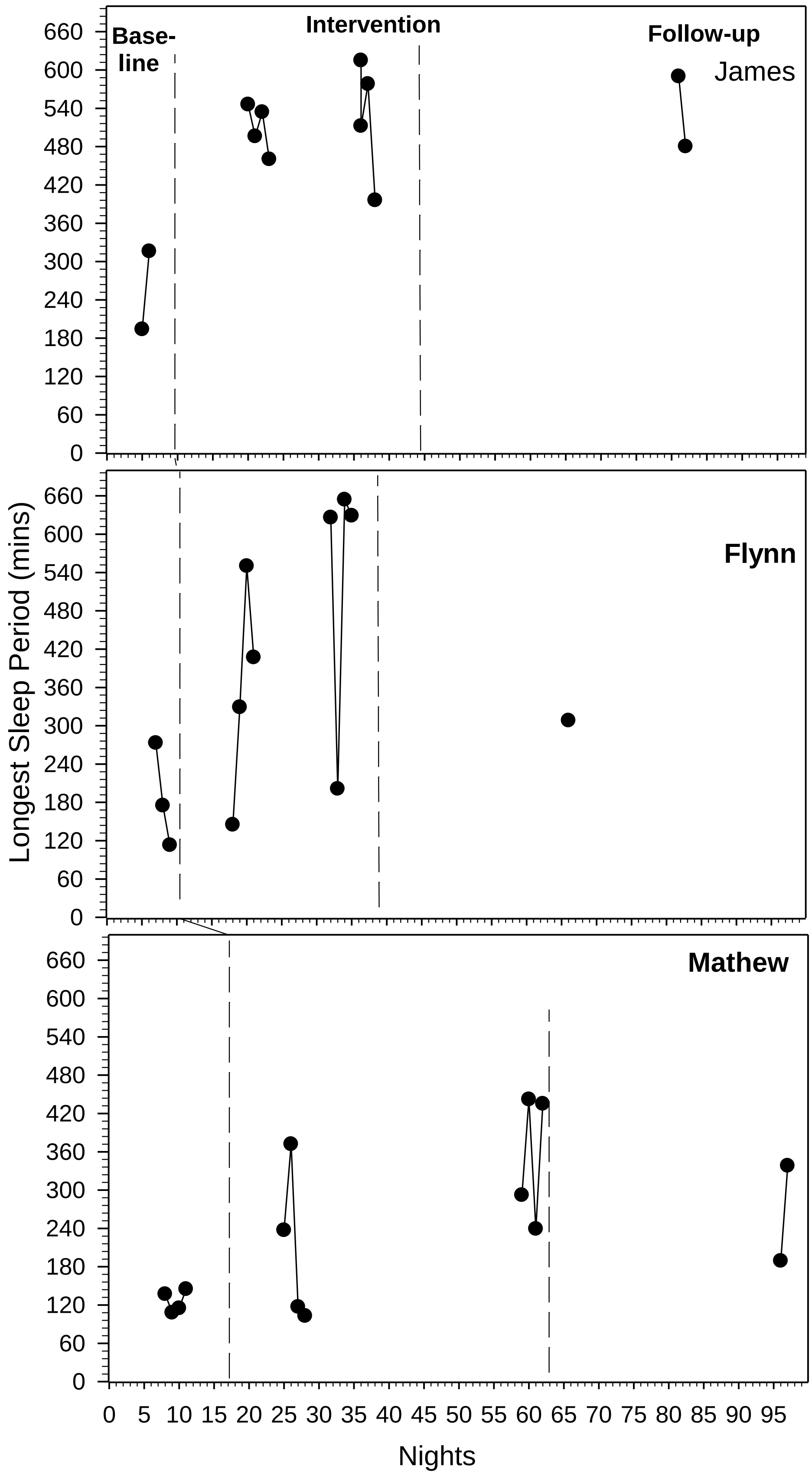

Figure 7. VSG data for Longest Sleep Period (LSP)

Sleep-Wake Transitions

Number of transitions from sleep to waking is illustrated in Figure 8 for all three participants. James experienced a dramatic decrease in the number of sleep-wake transitions from baseline (16 to 20 transitions) to intervention (two to four transitions). This reduction remained stable across intervention and follow-up. Flynn too experienced a marked decrease in sleep-wake transitions from the beginning of intervention with a brief recovery on night 66 at follow-up. During baseline, Matthew’s number of sleep-wake transitions range from 15 to 17 per night. During the start of intervention, Matthew’s transitions dramatically decreased to seven transitions. Across intervention, Matthew typically transitions between sleep and wake three to eight times a night, with the exception of one outlier (14) on the eleventh night of recording. The decrease in sleep-wake transitions remained stable at follow-up.

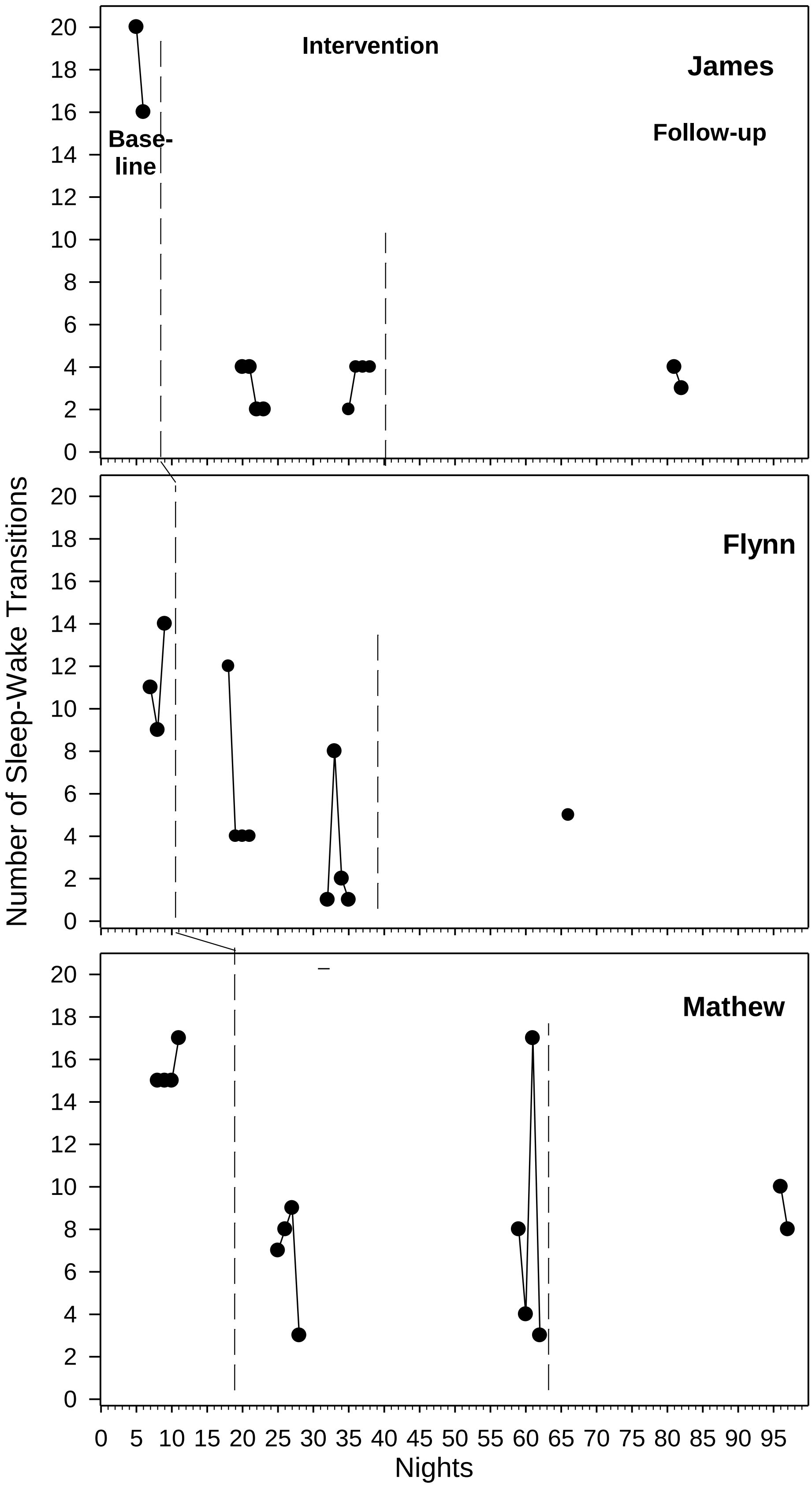

Figure 8. VSG data of number of sleep-wake transitions

Largest Active-Quiet Sleep-State Transitions before Waking

The Largest Number of Active-Quiet Sleep-State Transitions before Waking was measured across baseline, intervention and follow-up and is illustrated for each participant in Figure 9 as a measure of sleep continuity. In baseline all three infants sustained relatively few transitions before waking. For James, transitions increased to between 10 to 16 during intervention, with the exception of one night of disrupted sleep, and this change remained consistent at follow-up. Flynn's transitions at intervention steadily increased with the exception of one outlier near the end of intervention. By the end of intervention Flynn slept between 17 and 22 transitions without interruption before he woke. This change did not continue at follow-up and instead illustrates a decrease in transitions which coincided with earthquakes within the Canterbury region. Matthew's transitions at intervention varied, however, by the end of intervention increased to between 7 and 13. This change did not remain consistent at follow-up.

In summary, the data presented below illustrate similar trends to that noted with LSP. All three infants show an increase in the number of transitions before waking at intervention which suggests that they could sleep through a greater number of Active-Quiet sleep-state transitions before waking in the night or for the day.

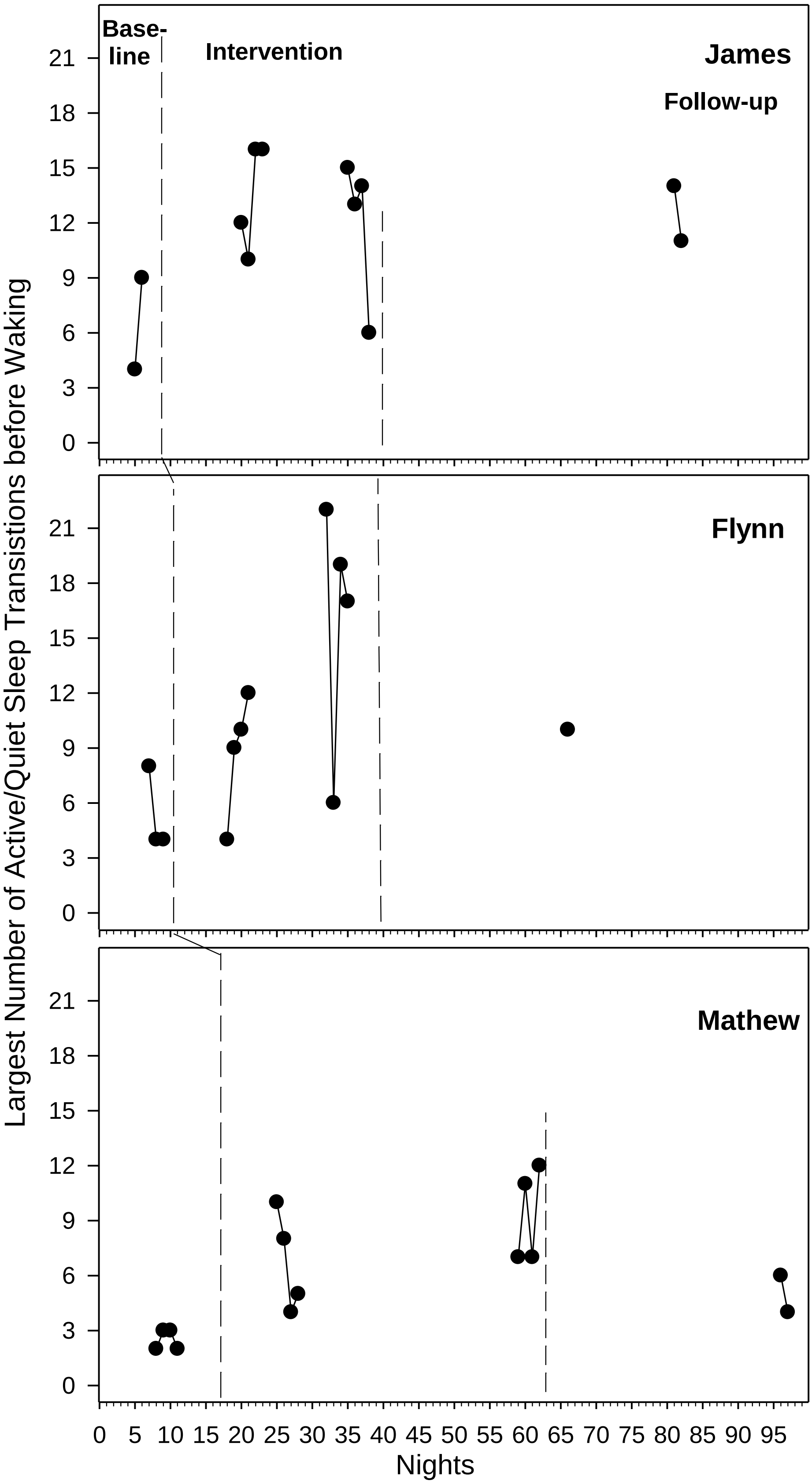

Figure 9. VSG data for largest Number Active-Quiet sleep-state transitions before waking

Parent Evaluation Questionnaire

All parents strongly agreed that the intervention was helpful in teaching them skills to manage their infant's sleep, that it was an effective programme and that they would recommend it to other parents. When responses were scored all parents fell within the high satisfaction/low stress category, with scores of 42 (James' mother), 40 (Matthew's mother), and 39 (Flynn's mother).

Discussion

The purpose of the current study was to implement an established behavioural sleep intervention, namely parental presence, with parents of infants who had sleep disturbance, in order to investigate the effects of a successful intervention on sleep-state organisation. Parental presence has been found to be effective in numerous studies (e.g., France & Blampied, 2005; Sadeh 1990, as cited in Sadeh & Anders 1993; Sadeh, 1994; Sadeh et al., 2010; Selim et al., 2006), but these studies have relied mostly on parent observation which does not detect silent aspects of sleep. While parental report indicates a decrease in sleep disturbance as measured by reductions in signalled night waking, VSG offers an opportunity to closely examine these features of sleep not detectable to parents such as sleep-state organisation and periods of quiet awake. The present research attempts to address whether an established behavioural sleep intervention changes infant sleep, resulting in an increase in healthy sleep behaviours (sleep-state organisation) or whether infants are still waking, but are not signalling to their parent/s.

Research Questions Revisited

To discuss the research findings in relation to the present research questions, it is first necessary to revisit whether infants and their parents met research criteria, by establishing whether a) infants in fact had ISD, and if so in what way, and b) whether parents adhered to the behavioural sleep intervention by changing the way they interacted with their child at bedtime and later in the night. Once this is established the impact of these changes in parent behaviour on infant sleep can be investigated by closely analysing a) infant's response to intervention and b) changes in sleep-state organisation. These are discussed in detail below.

Were Research Criteria Met?

Summary of Infant's Problematic Sleep

The degree of ISD was measured by calculating ISD scores (Healey et al., 2009). In baseline, James had the lowest ISD score indicating only a moderate level of ISD. Sleep Onset Delay (SOD) in particular was not problematic in that it was short but parents reported he required parental interaction at times to initiate sleep and sleep diaries stated on some occasions he was placed to bed once asleep. Hence, SOD, sleep duration, and duration of wakings (quiet and audible) did not change across the length of the study as would be expected for an infant who has acquired self-soothing skills and applied them to their sleep.

Flynn had more pronounced sleep difficulties and experienced problems within all areas of sleep, including, SOD, frequency of wakings, duration of wakings (audible), and Sleep Efficiency. Matthew also had many problematic sleep behaviours including, significant difficulties with SOD, frequency of night wakings, duration of night wakings, and Sleep Efficiency. Matthew also had the most significant ISD as determined by his composite ISD score and it should be noted that Matthew experiences both changes in his physical environment (i.e., parents bed to own bed) and the environmental change in parent-child interaction (i.e., parental presence procedure).

Parental Interaction

During intervention, parental interaction decreased to low levels and on most nights ceased, indicating that all parents adhered to the no-interaction aspect of the intervention. Parents were providing a range of interactions during baseline (i.e., cuddling, feeding, rocking) but these largely or completely ceased during intervention and maintenance, Changes in infant sleep during these phases have to be seen in the new context of very much reduced levels of parent interaction, with clear implications for the occurrence of self-soothing. In the absence of parental soothing self-soothing is assumed to have increased concomitant with the positive changes in infant sleep that are discussed next.

Effect of Intervention on Infant Sleep

Infants' Response to Intervention

Infants' responses to intervention were measured first by those variables that could be detected by their parent/s. Favourable outcomes of a sleep intervention include a decrease in SOD where appropriate, a reduction in the number of night wakings, and a reduction in the duration of audible night wakings. All three participants in the present study responded to treatment differently (especially in the rate of change displayed) but favourably, illustrating that the sleep programme had improved their sleep, as assessed by the three conventional measures, however sleep variables that changed were only those which were problematic for the infant prior to intervention. For instance, James' SOD and duration of crying was not regarded as problematic in terms of duration and remained relatively stable across each phase of the study. On the other hand the frequency of his night wakings was problematic and after intervention had improved. Both Flynn and Matthew also responded to treatment by decreasing their problematic sleep behaviours (SOD, night wakings, and duration of audible wakings) and increasing desirable sleep behaviours (nights of 'sleeping through').

Summary of Results

All three participants responded positively to the sleep programme, with nights of 'sleeping through' increasing during the course of the study. For James, his night wakings decreased as did his number of sleep-wake transitions. Flynn's SOD, number night wakings, and number of sleep-wake transitions decreased as intervention progressed. Duration of time spent awake, and time spent quietly awake increased during the start of intervention compared to baseline and then decreased during the second half of intervention. Matthew's SOD and sleep-wake transitions waxed and waned typically in response to some disruption. Each night of disrupted sleep however, reduced with an increase in nights of minimal SOD and sleeping through. His night wakings reduced as did the duration of time spent quietly awake. Duration of crying and time spent awake increased during the start of intervention and then steadily reduced.

Sleep-State Organisation

Sleep-state changes were measured by duration of awake, duration of quiet awake, number of sleep-wake transitions, the largest number of Active-Quiet Sleep-State Transitions before waking, and LSP. If a behavioural intervention does impact sleep-state organisation we would likely see a decrease in the duration of time spent awake and an increase in Sleep Efficiency. Furthermore, fewer sleep-wake and sleep-state transitions would indicate a maturing of sleep-state organisation. LSP, and Largest Number Active-Quiet Sleep-State transitions before waking should also increase, indicating an improved organisation of sleep, in response to intervention.

Alternatively, if self-soothing skills alone were influenced by the intervention, infants duration of time spent quietly awake would increase as the child switches from signalled awake to quiet awake states. Little change would be evident in other sleep-state variables from that during baseline. On the other hand, a combination of the two changes discussed may occur in which we would likely see changes over time in sleep-state organisation giving an indication of learning and consolidating self-soothing skills in combination with organising sleep-states as well as changes in quiet awake.

Duration of awake. Duration of time spent awake was categorised into quiet or audible awake both of which increased in duration at the start of intervention and decreased back to baseline levels mid-way through intervention. This trend is not unexpected as research does suggest that during extinction procedures an increase in problematic behaviour is likely to increase before it improves, in fact this is observed in most behaviour change and is referred to as Post Extinction Response Burst (PERB) (Lerman & Iwata, 1995). Lerman and Iwata (1995) found that of the studies reviewed involving extinction as an intervention, 24% reported a PERB and that this was most likely in those studies removing behavioural reinforcement for undesirable behaviour/s alone as an intervention. Within the present study, parental interaction for sleep initiation and night wakings is removed or reduced in both intervention and maintenance phases. Infants are therefore encouraged to resettle themselves to sleep upon night wakings which is likely to be difficult whilst establishing self-soothing skills.

Anders et al (1992) conducted a study which in part confirmed that infants at all ages wake in the night, however, by eight months of age they are more likely to wake without signalling than at three weeks of age. This suggests that when infants did wake, they spent a greater portion of time quietly awake than at the younger age. This is important in that it suggests changes in self-soothing skills. This finding was supported within the present study. Specifically, when comparing quiet with total duration of awake for the last few nights of intervention for Flynn and Matthew (as these were disruptive areas of sleep for them), the time spent quietly awake consumed the majority of total time awake. Hence, these infants while still spending minimal time awake per night tended to wake quietly rather than signal for their parents. This is important for the present research as it suggests that self-soothing skills were learnt over the intervention and were utilised upon infant night wakings.

The decrease in duration of time spent awake mid-way through intervention is likely to provide some evidence for the establishment of self-soothing skills. In other words, this suggests that prior to learning these skills, infants spent more time awake but that over the course of the study there was a period during intervention in which infants learnt self-soothing skills and by the end of intervention spent less or equal time quietly awake than during baseline. It would be unexpected to see large changes in the duration of awake by follow-up as parents are being signalled upon wakings during baseline and their parental interactions are likely to soothe infants to sleep in a timely manner. Parent interviews, sleep diary data and VSG data confirmed there was a significant difference between parent-child interactions during baseline compared to follow-up. Hence, the two time periods are measuring different variables and a desirable outcome may not necessary pertain to a decrease in duration of awake but rather the method of infants resumption to sleep. During baseline duration of infants night wakings (both quiet and crying) was found to be influenced by parents' interactions and the degree to which their parents could settle them. Typically parents in the present study were found to, rock, breastfeed, bottle-feed, or cuddle their infants upon their night wakings. During intervention parental interaction is limited and infants learnt to settle themselves by self-soothing. Hence, by follow-up, infants duration of awake is influenced by their ability to self-soothe back to sleep without parental intervention.

Sleep Efficiency. Sleep Efficiency scores illustrate the proportion of time infants spent asleep per night. A night is measured as the time the infant is placed in their cot until the moment they leave their cot, once they are up for the day. A high Sleep Efficiency score is favourable. While two of three participants showed an increase in Sleep Efficiency after intervention, the most notable change occurred during the intervention. Research suggests that infants with ISD typically rely on their parent/s to settle them to sleep (France & Blampied, 1999; Meijer, 2011). It is not surprising then that Sleep Efficiency dramatically decreases when parental response to night wakings is removed. In other words that infant's spent a greater proportion of their night awake and less time asleep during the initial stages of intervention.

During baseline infants' sleep was problematic and therefore it is likely that parents would attempt to keep them in their cot for a longer period to make up some of the hours of lost sleep. In other words, parents are likely to have kept their infants in their cots for a longer duration during baseline, compared to other phases of the study. This suggests that Sleep Efficiency scores alone are not accurately illustrating the changes in sleep after intervention. To investigate this further it is important to address the change in other aspects of sleep including, LSP and sleep-state transitions.

Longest Sleep Period (LSP). The LSP is a measurement of an infant's physiological capacity for continuous sleep (Anders et al, 1983). The present study found that there was a significant increase in LSP after intervention as did Hall and colleagues (Hall et al., 2006). This change suggests increasing maturity in sleep state-organisation. Infants in the present study slept for a longer duration, without waking, after intervention. From baseline to mid intervention, the increase was dramatic further suggesting that change exceeds that which would be typically seen in the natural trajectory of sleep-wake consolidation over the time period of this study.

Largest Active-Quiet sleep-state transitions before waking. Waking in the night is not a determinant of ISD, and in fact most infants wake when transitioning between Active and Quiet sleep (Henderson et al., 2011). The arousal level experienced and other events that occur during these wakings determines whether an infant becomes unsettled or can settle back to sleep. An infant with difficulties self-soothing is vulnerable to ISD as may the case for an infant who frequently transitions between sleep-states. In other words for infants with ISD, sleep-state transitions can precede waking. During baseline, infants in the present study experienced few sleep-state transitions before waking and were more likely to wake after a sleep-state transition. Interestingly, after intervention all three infants slept through notably more sleep-state transitions than during baseline. This, in combination with changes observed in LSP suggests that sleep became more organised indicating desirable changes in sleep-state organisation. Furthermore, when they did wake, infants were more likely to soothe themselves back to sleep illustrating both changes in sleep-state organisation and self-soothing or management of night wakings. LSRSP was not illustrated in the present results. It was determined the LSRSP would similarly increase as its components (LSP and duration of quiet awake) did, however, in light of Henderson et al 2010 it is recommended that future research concentrates on investigating this measure, especially given the present results indicate a notable change in sleep-sate consolidation and the LSRSP is considered an ideal measure to capture this change.

Limitations of the Current Study

One limitation of the current study was that the videos were viewed and coded at the end of the study. The process of coding was time consuming and hence the frequency and number of video recordings were limited for each participant. Consequently, a stable baseline was not established using the VSG data for James. Only two nights of recording were viewed for James at baseline. On one of these he was likely to be suffering and/or recovering from a cold and this somewhat constrained the interpretation of his VSG data as there was insufficient baseline data to detect trends. It would have been valuable to extend the baseline recordings for James, and indeed for all the participants, if technical and resource difficulties could have been overcome.

Unforeseen environmental events affected data collection and parts of data analysis. While the most significant earthquakes occurred prior to the study, continual aftershocks did disrupt some aspects of daily living, including power cuts and parental stress. At the time of data collection, Canterbury residents experienced a series of earthquakes and/or aftershocks. Due to the nature of data collection, visual recordings were vulnerable to technical failures, including damaged equipment and power cuts. This meant that some hours of visual recording, and on some occasions entire nights, were lost. The placement of the video equipment required particular attention so that if in the event of a significant earthquake, where equipment is likely to fall or move, the infant is away from this foreseen hazard. It was made clear to each family that in the event of an earthquake and/or aftershock that they should respond accordingly by ensuring safety of their family members. It was also important to communicate regularly with parents to ascertain whether infants woke during aftershocks and if they were fearful so that accommodations could be made if necessary, which may have included cessation of the study until an appropriate time. This was not necessary for any infants and in fact with the exception to Flynn's response during follow-up, all were reported to be unaware of the earthquakes and/or aftershocks. Furthermore, each family reported feeling comfortable with administering the intervention at this time.

The natural trajectory of infant development typically illustrates a consolidation of sleep-wake and sleep-state organisation over time. Efforts were made to control for this as much as possible by applying a multiple-baseline-across-participants whereby participants act as their own control. This design involves applying intervention at different time periods for varying lengths of times. The results indicated that changes in many sleep behaviours occurred at intervention phase for all three participants, suggesting an intervention rather than developmental effect. Furthermore, participants were of different ages and trends were noted to be rapid rather than unidirectional (which is what would be expected in development). The length of the study from start to finish was also relatively short and from this, in combination with that stated above, it can be inferred that intervention outpaced natural maturation of sleep-state organisation.

Implications of the Current Study

The present study aimed to address the nature of change that occurs when changing a feature of the infant's proximal environment assumed to engage security and attachment mechanisms while concurrently changing parent responses to waking as happens in the parental presence procedure when used for infants with ISD. The present findings suggest that intervention aids in improving infant sleep within areas that are problematic in the first instance. Measurable change noted in the present study included sleep-state consolidation (increase in LSP and Active-Quiet sleep-state transitions before waking), a likely increase in self-soothing behaviours (changes over time in Duration of Quiet Awake and decreases in parental interaction) and sleep consolidation (decrease in SOD, night wakings, and sleep-wake transitions).

Evidence that sleep-state organisation is impacted by sleep intervention was supported in the present study. In particular it was noted that infants were able to sleep for a longer period before waking and that they sleep through a greater number of Active-Quiet sleep-state transitions before waking after intervention. Implications support the idea that sleep environmental changes that promote consistency and self-initiation like those seen when removing excessive parental interactions at night can positively change the organisation of sleep itself.

The change observed for the two infants who had settling difficulties prior to intervention also provides evidence of the positive effect of parental presence on SOD. These two participants displayed a dramatic increase and then decrease to levels equal to or below baseline in duration of time spent awake and quiet. As outlined earlier this likely suggests changes in self-soothing behaviours which are important for current research and practice. Self-soothing behaviours are also an important component of emotional regulation and the effects of those changes in self-soothing illustrated in the present study, while not yet evident, are likely to generalise to other areas. Future research should focus on investigating this further and by specifically analysing visual aspects of self-soothing. The present study assumed that periods of quiet awake were indicative of self-soothing; however this may not always be the case. Visual evidence of self-soothing could include, thumb sucking, cuddling toys/blankets, using a pacifier, or twiddling blankets or hair. These behaviours were observed in the present study (VSG data), however, were not differentiated from periods of awake and quiet (without visual self-soothing).

Collectively these findings also provide a detailed model of sleep behaviour change that could be used to measure effectiveness of infant sleep intervention. While this is the beginning of research addressing this question, further research within this area may in practice allow professionals who implement sleep interventions to promote the benefits of sleep intervention on sleep-state organisation and self-soothing behaviours. This further supports the idea that environmental changes including changing parental interactions at night like those implemented in the present research can positively influence the trajectory of sleep development.

It is therefore recommended that further research of this kind is conducted. Furthermore, the present research raised the question of different sleep-state organisation profiles and their impact on ISD. In other words, if as this study suggests, an improvement in sleep is associated with changes in sleep-state organisation, do infants with severe ISD have different sleep-state organisations relative to those who had less significant ISD, and furthermore, those who have developed healthy sleep without intervention.

Conclusion

The current study found that an established intervention for ISD that encouraged self-soothing and routines and extinguished or reduced parental interaction did improve sleep outcomes for all infants. Most notable were changes in sleep-state consolidation which included an increase in LSP and the number of Active-Quiet sleep-state transitions before waking. Furthermore the changes noted in the duration of time spent awake and quiet and the observation that infants achieved desirable levels of sleep behaviour on their own suggest an important role for self-soothing. This suggests that behavioural sleep interventions implemented for infants can increase their self-soothing which results in less signalling (i.e., crying). During the initial stages of intervention these skills were perhaps weaker and infants spent a greater duration of time awake (including both audible and quiet). As intervention progressed, changes in the duration of quiet sleep were noted. For a short period of time infants appeared to be lying awake at equal or greater durations to that during baseline, however over time, once self-soothing skills were consolidated, infants spent little time awake at night and when they were awake they were likely quiet. This suggests that environmental changes had a direct impact on sleep (which may have been mediated through self-soothing skills) by increasing the amount of time infants could sleep before waking and increasing the number of transitions they could sleep through before waking. Rather than the relationship between sleep intervention and self-soothing or sleep-state consolidation, being either or, the effect appears to involve changes within both, creating the cascading effect discussed within Sadeh and Anders (1993) model of infant sleep.

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Appendix A

Information sheets

Canterbury Sleep Programme:
Psychology Department and
Health Sciences Centre

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

<u>Information Sheet for Primary health Organisation</u>
Family intervention
for infant sleep problems

The Canterbury Sleep Programme is calling for referrals of infants or toddlers aged 6-24 months who have difficulty settling to sleep or who wake frequently at night. Families will receive a free assessment and a free supervised, tailored sleep programme. In return, families will be asked to record their infants sleep prior to and during the programme and to allow a low illumination camera to be set up in their baby's room for up to eighteen nights (approximately 2-4 nights before intervention, 8 nights during the behavioural intervention, and 2-4 nights after the intervention).

This research is suitable for families who are able to wait up to one month before beginning a sleep programme. It is not suitable for infants with medical conditions likely to affect sleep (such as asthma), infants with severe family dysfunction making a sleep programme unwise, or infants with a developmental delay.

Clinical work will be carried out and/or supervised by Dr Karyn France, Registered Clinical Psychologist and Principal Investigator of the Canterbury Sleep Programme.

My name is Shannae Wilson, and I will be responsible for collecting data and analysing that data. I am a student at the University of Canterbury who is currently studying towards a Postgraduate Diploma in Child and Family Psychology and a Master of Science, majoring in Psychology. As part of my current course I am required to carry out a thesis. I have chosen to work within the Canterbury Sleep Programme to study changes in the sleep of young children undergoing an intervention to help with their sleep problems.

There will be no charge for the assessment and intervention which will be delivered by me together with Dr Karyn France.

As part of the completion of my Masters qualification a final write-up will be accessible as a public document via the University of Canterbury in the library catalogue. The final report may also be published, and in both instances all identifying information will be withheld. No images will be included in the final document; therefore, parents can be assured that no one reading the final report will be able to identify them or their child.

To refer families please contact me at any time on 027 3242 426 or you can email me at slw89@uclive.ac.nz.

If you have any questions please feel free to contact either of my supervisors, listed below.

Kind Regards,

Shannae Wilson

Dr Karyn G France,
Registered Clinical Psychologist,
Coordinator Child and Family Psychology Programme
University of Canterbury
Private Bag 4800,
Christchurch, New Zealand.
Ph (03) 364 2610   Fax (03) 364241

Neville M Blampied
Head of Department of Psychology
University of Canterbury
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Canterbury Sleep Programme:
Psychology Department and
Health Sciences Centre

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Information Sheet
Family intervention
for infant sleep problems

To the parents/Caregivers

My name is Shannae Wilson, I am a student at the University of Canterbury who is currently studying towards a Postgraduate Diploma in Child and Family Psychology and a Masters in Science, majoring in Psychology. As part of my current course I am required to carry out a thesis. I have chosen to work within the Canterbury Sleep Programme to study changes in the sleep of young children undergoing an intervention to help with their sleep problems.

If you have an infant or toddler aged 6-24 months who has difficulty settling to sleep or who wakes frequently at night you may be eligible to take part. If this describes your child you are invited to make contact with me.

There will be a short interview on the telephone to hear a little about your child and their sleep and to answer your questions about the project. If we both agree then this will be followed by a longer interview at our clinic to gather full information about your child and their sleep and to explain possible interventions.

The aim of the research is to reduce sleep disturbance in infants who currently experience them. In order to do this, a behavioural family intervention will be explored. This will be a personalised sleep programme and the details of this programme will be negotiated with you throughout the entire process.

There will be no charge for the assessment and intervention which will be delivered by me together with Dr Karyn France, Registered Clinical Psychologist and Principal Investigator of the Canterbury Sleep Programme.

There will be a brief baseline phase during which we will record your child's sleep as it is now, followed by a sleep programme negotiated with Dr France. In return we ask to record your child's sleep overnight for approximately 18 nights over a 20 week period using a low-illumination camera. The use of this technology in homes is well established in the child sleep field. The camera and supporting equipment will be placed in a safe position which will be negotiated with you. Only the child and their bed will be visible, but background noise will be able to be heard. I will set up the equipment and programme it to start and finish at set times. This will not require anyone else entering your home at night.

The nature of this study is such that we need to record your child's sleep as it changes over several days at a time. We anticipate that recording will occur on two nights at the beginning of the programme, eight nights during the sleep programme and a further two nights during follow-up.

Throughout the programme we will also ask you to complete a nightly sleep diary. We will also ask you to complete a questionnaire at the end about how you found the study.

The sleep programme will be negotiated with you and it will be a well-established programme noted to be as gentle as possible.

Sleep programmes, have been demonstrated to be effective in managing sleep disturbance in infants. The Canterbury sleep programme has worked over many years to establish which sleep programmes are the most gentle, while also being effective. This will be thoroughly discussed with you by Dr France, prior to your agreeing to follow a sleep programme. You will be contacted regularly as you carry out the programme, so we can answer any questions and help you to solve any problems which may occur.

Although we have good data on the effectiveness of sleep programmes we have little information on how children's sleep reorganises during the programme, hence the need for sustained video recording during the intervention.

The video recording is to obtain information on your child's sleeping behaviours, specifically; how long it takes them to fall asleep, the proportion of REM (active) and nonREM (quiet) sleep they experience, the number of times they wake during the night, and the length of time it takes for them to reinitiate sleep. This information is gathered from the videos, back at the university, using a standardised coding system.

In addition, we will ask you to record brief information in a sleep diary that will be provided for you. This is a two page form that has space for recording seven nights of sleep and has room for you to record times your child goes to bed, and to sleep, awakens during the night and finally in the morning. It also asks you to record what you do to try to get your child to sleep.

It is envisioned that this research will require approximately eight home visits; these visits will be brief and only take approximately 5-10 minutes with the exception of the first which may take between 15-30 minutes. These visits will allow me time to set up or collect the video equipment and also allow us time to touch base and discuss progress. These visits are necessary to ensure the correct application of the programme is being applied and to provide support and advice where you may need it. You will also be asked to spend a short time filling in sleep diaries, questionnaires and switching on and off the camera.

All identifying information will be kept confidential between myself and my two research supervisors. As part of the completion of my Masters qualification a final write-up will be accessible as a public document via the University of Canterbury in the library catalogue. The final report may also be published, and in both instances all identifying information will be withheld. No images will be included in the final document; therefore, you can be assured that no one reading the final report will be able to identify you or your child.

Should you and your child decide to participate in the study, you have the right to withdraw at any time without reason or without it affecting the way you are treated. If you choose to withdrawal your infant, any data that was collected until this point will be discarded. Throughout the entire research you also have the right to ask questions and voice concerns.

This is also an opportunity to receive free professional assistance, advice, and work with us in using some of the leading interventions used to manage sleep problems in young children.

The interventions will be negotiated with you in a professional manner and you will be well-supported throughout. The project has been reviewed and approved by the University of Canterbury Human Ethics Committee and there are no known risks or adverse side effects to the interventions that will be implemented.

Thank you for taking the time to consider this information sheet. If you require any further information please do not hesitate to contact either of my supervisors as listed below.

Kind Regards,

Shannae Wilson

Shannae Wilson
Ph 027 3242 426
Email: slw89@uclive.ac.nz

Dr Karyn G France,
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Appendix B

Semi structured interview questions

1) Tell me about your child's sleep now?

2) What is a typical night like in your house?

3) What time do you usually place your infant in their cot or bed?

4) How long after you place your infant in their cot or bed do they settle to sleep?

5) During a typical night, how many times will your infant wake?

6) What time will your infant typically wake up in the morning?

7) How do you handle the sleep problem now?

8) What have you tried in the past?
    a. How did that work out?

9) Has ***infant's name*** been put on any medication for their sleep problem?

10) When did you first notice that ***he/she*** had a sleep problem?
    a. Was there a clear reason for the sleep problem to begin?

11) Has ***infant's name*** sleep problem been continuous or was there a period of settled sleep?

I am now going to ask about ***infant's name*** development so far:

12) How were things for you as a family when you knew ***he/she*** was coming along?

13) How was your pregnancy with ***infant's name***?

14) How was your birth with ***infant's name***?

15) Tell me about ***infant's name*** as a very new baby?
    a. ***His/her*** crying?
    b. ***His/her*** activity?
    c. ***His/her*** sleep?

16) How did ***he/she*** take to feeding?
    a. Tell me about that?

17) Tell me about ***infant's name development (this will vary depending on infant's age and observations prior to questionning-e.g is the infant crawling yet?).***
    a. What age did ***he/she*** start crawling?
    b. What age did ***he/she*** start walking?
    c. What age did ***he/she*** start talking?

18) Has ***infant's name*** experienced any illness?

19) How is ***his/her*** health now?

20) What have there been in the way of ups and downs or changes for the family over ***infant's name's*** life so far?

21) Have you any other concerns about ***infant's name or anyone else in the family***?

Appendix C

Sleep diaries

Child's Name: ______________________________Week:_____________________________

Condition/Phase of Study: ____________________ Goal Bedtime: ____________________

Day Sleep							
	Please insert the date in row below						
	___/___/___	___/___/___	___/___/___	___/___/___	___/___/___	___/___/___	___/___/___
Time put in bed/cot:							
Time infant wakes up:							
Where is infant sleeping:							

Night Sleep							
Please insert the date in row below							
	___/___/__	___/___/___	___/___/___	___/___/___	___/___/__	___/___/___	___/___/__
Where is the infant initially put to sleep for the night:							
Actual bedtime:							
Ideal bedtime:							
Minutes from in bed to silence (first time):							
If wakes, please briefly describe noise infant makes:							
Time the infant wakes up (for the day):							
Where the infant wakes from (I.e. their cot, bed, your bed etc):							
Comments:							

Night Sleep Cont.

	Please tick boxes for each time the infant wakes and note beside the tick how long they are awake for at each period and what you did after each awakening.									
(Insert Date)	1	2	3	4	5	6	7	8	9	10
___/___/___										
___/___/___										
___/___/___										
___/___/___										
___/___/___										
___/___/___										
___/___/___										

Appendix D

Parent Evaluation Questionnaire

Treatment Evaluation Questionnaire – Behavioural Family Intervention

(Adapted from Kazdin, 1980; Kelly, Heffer, Gresham & Elliot, 1989)

Please respond to each of the statements below by ticking an appropriate box that ranges from strongly agree to strongly disagree. Please make sure that each statement has a response and note that all statements are referring to only the behavioural intervention as a form of treatment.

	Strongly Disagree	Disagree	Neutral	Agree	Strongly Agree
1. I found the behavioural intervention to be an acceptable way of dealing with my child's sleep disturbance.					
2. The behavioural intervention has given me ideas and skills for continuing to manage my infant's sleep.					
3. I believe that it is acceptable to use the behavioural intervention with a young child.					
4. I liked using the behavioural intervention for my infant.					
5. I believe the behavioural intervention is an effective treatment for my infants sleep disturbance.					
6. I believe my child experienced discomfort during the behavioural intervention.					
7. I believe the behavioural intervention is likely to result in permanent improvement in my child's sleep.					
8. Overall I have a positive reaction to the behavioural intervention.					
9. I would recommend this intervention for sleep problems in infants to other parents.					

Thank you for taking the time to complete this questionnaire.

Appendix E

Human Ethics letters of approval

Ref: HEC 2010/133

29 September 2010

Shannae Wilson
Department of Psychology
UNIVERSITY OF CANTERBURY

Dear Shannae

The Human Ethics Committee advises that your research proposal "Managing infant sleep disturbance with Merino sleepwear followed by a behavioural intervention: effects on sleep-state architecture" has been considered and approved.

Please note that this approval is subject to the incorporation of the amendments you have provided in your email of 27 September 2010.

Best wishes for your project.

Yours sincerely

Dr Michael Grimshaw
Chair, Human Ethics Committee

Appendix F

Participant consent forms

Canterbury Sleep Programme:
Psychology Department and
Health Sciences Centre

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Consent Form
Family intervention
for infant sleep problems

I/we have read and understood the attached information sheet, and I/we have been given the opportunity to ask questions. From this, I/we understand what is involved for my/our child and for me/us.

I/We understand that I/we will not be responsible for any of the equipment and that the researcher/s will take care of all the equipment and set up involved.

I/We understand that all the information about me/us, my child and my family will remain confidential and that all written documents will exclude any identifying information. Identifying information will only be available to the participant, myself, the supervisors and the researcher.

I/We agree and consent to my/our child taking part in the study as outlined in the information sheet.

I/We understand that in signing this consent form I/we am consenting on behalf of my child and that I/we may withdraw my/our child at any time without giving reason or affecting the way I am/we are treated.

I/We note that this project has been reviewed and approved by the University of Canterbury Human Ethics Committee.

I/we____ (Please print name/s) agree for myself/us and ____ (Print child's name) to take part in the sleep study described in the attached information sheet.

Signature/s____Date________________